

TESTING FOR HEREDITARY CANCER PREDISPOSITION: THE IMPACT OF THE
NUMBER OF OPTIONS AND A PROVIDER RECOMMENDATION ON DECISION-
MAKING OUTCOMES

by
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ABSTRACT

Objective:

To examine how the presentation of a decision can influence choices about genetic testing for inherited cancer predispositions. Specifically, how the number of options and the addition of a personalized recommendation might influence outcomes such as the likelihood of undergoing genetic testing, the genetic test chosen, and whether a person's test choice matches their personal preferences.

Methods:

An online hypothetical vignette study was completed by 454 healthy volunteers. Each participant was randomized to receive one of two survey versions which differed in the manner of presenting testing options and how these options were integrated with a provider recommendation. Regression analyses were performed to determine the relationships between the presentation of choice and participant decisions. Wilcoxon rank-sign tests were used to determine the impact of a provider recommendation on final genetic testing choices.

Results:

Participants were more likely to choose to undergo genetic testing when presented with three options instead of two (OR: 2.00 $p=0.014$). This effect was no longer observed when individuals who had decided not to undergo testing were presented with a third option (OR: 0.90 $p=0.775$). The addition of a provider recommendation did not significantly change the overall distribution of options chosen ($p=0.746$). However, after a recommendation, participants were more likely to choose the test that best matched with personal preferences about the type of genetic information desired ($p<0.001$).

Conclusions:

Participants are more likely to undergo genetic testing when presented with more options.

They are also more likely to select an option in line with a personal preference if

presented with a recommendation based on this preference.

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PART 1: INTRODUCTION

ORGANIZATION OF THESIS

Part 1: Contains an overview of the organization of the thesis, an executive summary, the thesis objective and aims, a comprehensive literature review, and a detailed review of the methods

Part 2: Is a manuscript that focuses on the three thesis aims. Analytic methods and results for each specific aim are not addressed in this thesis, as the option to write a manuscript was selected. Additional results will be published in future manuscripts.

EXECUTIVE SUMMARY

Next-generation sequencing technologies have enabled the proliferation of panel genetic tests for inherited cancer predispositions. The growing number of testing options has made it more challenging for healthcare providers to decide which tests to offer and how to present them to patients because there may be multiple medically appropriate options. How providers present decisions is important because it can influence the choices that patients make and their subsequent medical care (Thaler & Sunstein, 2009). The purpose of this study is to examine how the presentation of genetic testing might influence whether people decide to undergo genetic testing and the specific test they may choose. Understanding how the presentation of genetic testing influences decisions is the first step toward helping providers build an evidence-based practice for presenting decisions in a way that maximizes client understanding and engagement in the decision-making process. This study seeks to address the three aims outlined below.

The first aim is to determine the impact of adding a third option on the decision to have genetic testing or not. Research in other fields has shown that adding additional

options can influence the selection of preexisting options (Tversky & Kahneman, 1974). This is believed to occur because adding an additional point of comparison can change how people evaluate options. The specific way in which adding another option influences choice depends on the relative attractiveness of the options. At this time, it is unknown how people view the relative attractiveness of different genetic testing options and how they weigh tradeoffs in the decision-making process. This study seeks to determine if individuals are more or less likely to undergo genetic testing when presented with an additional genetic panel test.

The second aim is to examine the impact of a provider recommendation on genetic testing choice. Previous studies have indicated that many individuals would like a recommendation about genetic testing (Geller & Bernhardt, 1998; Vadaparampil, McIntyre, & Quinn, 2010). This study uses survey logic to give each participant who chose to undergo genetic testing a recommendation for one of the two genetic tests and the opportunity to change their previous decision. Each recommendation is based on the participants answer to a question about whether they would prefer to learn only clinically actionable genetic information or all possible genetic information. The impact of the recommendation is determined by examining whether the distribution of tests chosen differs before and after the recommendation. Additionally, this study investigates whether the test selected is more likely to match the individual's information preference after the targeted recommendation.

The third aim is to determine if the overall method of offering testing, which integrates the complete process of presenting genetic testing options and a provider recommendation, influences outcomes. These outcomes include the overall distribution

of which genetic testing options are chosen, and the likelihood of undergoing testing. Furthermore, because adding another option increases the amount of information that decision makers need to process and therefore the complexity of the decision, this study examines if the overall method of genetic testing relates to patient reports of decisional conflict.

OBJECTIVE AND SPECIFIC AIMS

Objective: To examine the impact of the number of options and a provider recommendation on a hypothetical decision about genetic testing for hereditary cancer predisposition.

Aim 1: To examine the impact of adding a third genetic testing option on the likelihood of having genetic testing.

Question 1.1: What is the impact of adding a third option on the decision to have genetic testing or not?

Sub-question 1.1: Would the outcome change if non-testers who were presented with two options are offered a third option after they have made their initial genetic testing choice?

Aim 2: To examine the impact of a provider recommendation on genetic testing choice.

Question 2.1: Does genetic test choice differ after a personalized recommendation?

Question 2.2: Does a provider recommendation influence the likelihood that the final test choice matches the participant's information preference and the corresponding provider recommendation among testers?

Question 2.3: Does the test presentation method influence the likelihood that the final test choice matches the participant's information preference and the corresponding provider recommendation among testers?

Aim 3: To examine the relationship between the overall method of testing presentation, the choices made and decisional conflict.

Question 3.1: Does the genetic testing presentation method impact the final genetic testing option chosen?

Question 3.2: Does the genetic testing presentation method impact the final likelihood of undergoing testing?

Question 3.3: Does test offering method relate to decisional conflict and is this relationship mediated by information preference/test choice concordance?

LITERATURE REVIEW

Conceptual Framework

The objective of this study is to investigate if altering the presentation of genetic testing decisions can influence the choices made. Specifically, this study seeks to investigate if manipulating the number of testing options offered and adding a provider recommendation can influence patient decisions within the context of hypothetical hereditary cancer predisposition testing. These are characteristics of a healthcare encounter that a provider is likely to vary -- either purposively or not. By better understanding how the presentation of a choice may influence the process of patient decision-making we can more effectively help patients to make decisions more closely aligned with their preferences.

The conceptual model underlying this investigation is based on a decision-making model that proposes three main categories of factors that influence choice (Figure 1). These factors are divided into attributes of the problem, person and social context of the decision (Payne, 1993). Specifically, this study focuses on examining the impact of problem-related attributes on genetic test choice. Problem-related attributes are intrinsic aspects of the structure and presentation of a choice and can be divided into task and context components. Task effects describe structural characteristics of the decision problem, for example, method of response, number of outcomes, number of alternatives, time pressures, and information display (Payne, 1993). Context components include factors associated with the value of attributes in a specific decision context, including the similarity and overall attractiveness of alternatives. The second category of factors, person-variables, are attributes of the decision maker, such as the decision maker's cognitive ability and prior knowledge (Payne, 1993). Some person-variables are also measured as a part of this study.

While the social context within which a decision is made also influences decision making, this is not examined in this investigation.

Introduction to Hereditary Cancer Syndromes

Cancer is the second leading cause of death in the United States, surpassed only by Heart Disease (Hoyert & Xu, 2011). In fact, 1 in 2 men and 1 in 3 women in the United States will develop cancer at some point in their lifetimes (American Cancer Society, 2013). Cancer is composed of abnormal cells that develop different biochemical properties that confer enhanced abilities to proliferate and survive. They acquire these biochemical attributes through a series of genetic and/or epigenetic changes that occur in

a way that gives that cell a competitive advantage over neighboring cells (Schneider, 2012). Some individuals are born with a genetic mutation that places them at an increased lifetime risk to develop specific types of cancer, often at younger ages than people in the general population. These individuals are said to have a hereditary cancer syndrome, and it is believed that about 5-10% of all cancers are inherited via this mechanism (Garber & Offit, 2005). There are at least 45 identified syndromes with clear genetic causes that confer an increased lifetime risk of developing cancer (Riley et al., 2012). These syndromes can differ on many dimensions including the types of cancer, the magnitude of the risks, the inheritance pattern, and whether individuals have other physical symptoms. Some hereditary cancer syndromes have been well described and studied for many years, while others are poorly understood.

Many types of cancer are associated with hereditary syndromes including breast, colon, ovarian, pancreatic, and kidney cancers (Schneider, 2012). Two of the most common hereditary cancer syndromes are Hereditary Breast and Ovarian Cancer Syndrome (HBOC) and Lynch Syndrome also known as Hereditary Non-Polyposis Colorectal Cancer syndrome. HBOC is caused by a deleterious mutation in either the *BRCA1* or *BRCA2* gene (Garber & Offit, 2005). This syndrome is associated with high lifetime risks of developing certain cancers which exceed general population risk including up to an 80% lifetime chance to develop female breast cancer, 40% chance to develop ovarian cancer, and elevated risks for prostate and pancreatic cancers (Petrucelli, Daly & Feldman, 2013). Approximately 1 in 800 women in the general population and 1 in 40 women of Ashkenazi Jewish ancestry have this syndrome (Schneider, 2012). In contrast, Lynch syndrome can be caused by a mutation in one of six different genes and

increases the chance of developing colorectal, endometrial, ovarian, small bowel, hepatobiliary, and urinary tract cancers. Most hereditary cancer syndromes including HBOC and Lynch Syndrome are inherited in an autosomal dominant manner; however, there are several syndromes which are inherited in an autosomal recessive manner, x-linked recessive manner or via parental imprinting disorders (Schneider, 2012).

Genetic Testing for Hereditary Cancer Syndromes

Genetic testing on a blood or saliva sample can be used to identify individuals with hereditary cancer syndromes and inform their cancer risk assessments. Individuals identified at elevated risks for cancer by genetic testing may use these results to guide medical management decisions about cancer treatment, screening and/or prevention with the objective of reducing cancer morbidity and mortality. For example, women identified as having HBOC may decide to have increased breast surveillance or have prophylactic surgeries such as a bilateral mastectomy or salpingo-oophorectomy due to their high risk of breast and ovarian cancers. Genetic test results can also be useful for informing cancer risks and guiding genetic testing in relatives. However, not all information from genetic testing is considered clinically actionable. For example, it is possible to learn about risks for cancers that cannot be screened for or prevented. Genetic tests can also yield results that are uninformative or unclear. For instance, a negative test result in the absence of a known mutation in a family does not indicate a decreased risk of cancer. Additionally, some genetic changes are not understood well enough to be classified as disease causing or not. These genetic changes are called variants of uncertain significance.

Typically, a person would come to clinical attention as a candidate for hereditary cancer testing based on a notable family or personal history of cancer or if a mutation is

identified in a relative. Elements suggestive of a hereditary cancer syndrome in the family include a family history with many cases of an uncommon cancer, cancer occurring at younger ages, multiple family members with a certain constellation of cancers, or a person with multiple primary cancers (American Cancer Society, 2013). In cases where the client sees a genetic counselor, the counselor would perform a risk assessment based on the client's personal medical information and family history of cancer (Riley et al., 2012). This risk assessment would serve to determine whether the client could be considered to have an average, modest, or increased cancer risk using a subjective assessment and sometimes empirical cancer risk models (Riley et al., 2012). The counselor would then determine whether the client is a candidate for genetic testing, and if so decide which genetic tests to offer and how to offer them. Clients who are offered testing then need to make a decision about whether or not to have genetic testing and potentially which test they would prefer to have.

These decisions have become more complex over time as new technologies such as next-generation sequencing have enabled the sequencing of multiple genes simultaneously (Hall, Forman, & Pilarski, 2014). Consequently, it is possible to test multiple genes on a single panel test and as a result many companies have begun to offer a variety of panel tests that have opened up new approaches to genetic testing for cancer risk assessment. At this time there are 4 general approaches to cancer genetic testing including the following:

1. Syndrome specific test (e.g. testing for HBOC)
2. Cancer-specific high penetrance gene panel (e.g. genes for several syndromes that cause a high-risk for breast cancer)

3. Cancer-specific gene panel with high and moderate penetrance genes (e.g. many genes that have some association with increased breast cancer risk)
4. “Comprehensive” cancer panels that include genes associated with multiple cancers or hereditary cancer syndromes (e.g. genes associated with HBOC, Lynch Syndrome and other cancer syndromes) (Hall et al., 2014).

Each of these approaches has distinct clinically relevant advantages and disadvantages which constitute trade-offs in choosing one method over another. For example, testing a larger number of genes at a time may be more cost-effective, and may improve detection of cancer susceptibility mutations. However, this approach can also lead to an increased likelihood of finding mutations in genes without evidence-based management strategies, receiving genetic variants of uncertain significance, and have a longer testing turn-around time (Hall et al., 2014). Given these challenges, National Comprehensive Cancer Network (NCCN) recommends that panels only be offered in the context of a consultation with a cancer genetics professional (NCCN, 2014).

The Decision Making Process and Outcomes

Because of the complexity of the tradeoffs, NCCN recommends that the benefits, limitations and management recommendations be reviewed by the provider and discussed with patients prior to ordering a multi-gene panel (NCCN, 2014). The National Society of Genetic Counselors (NSGC) recommends that informed consent for genetic testing should include a discussion of the precise gene(s) being tested, the possible outcomes of testing, medical management issues specific to the test results, a review of the possible benefits, risks and limitations, and alternatives to genetic testing (Riley et al., 2012). As the number of genes being testing increases, informed consent using this model is not

feasible, as cancer panels may include only a few genes or as many as 49 different genes (Hall et al., 2014).

There are multiple approaches to studying medical decision-making. An investigator can decide to measure different aspects of the process such as the elements surrounding the task of decision making (e.g. role preference), the decision making process (e.g. deliberation), the decision itself, or the patient's outcome post decision making (Scholl et al., 2011). Additionally, a distinction can be drawn between observational measures of the competence of the parties involved in the decision and measures of the perception of the patient or clinician (Scholl et al., 2011). However, there is debate as to what constitutes a "good" decision and how the aforementioned attributes integrate into this assessment (Elwyn & Miron-Shatz, 2010).

One study was conducted to elucidate the decision making process for decisions about predictive genetic testing used a simulated decision task. The 20 participants in this study ranged in age from 18-34 years old, had no family history of a known genetic condition with available predictive testing, and had not considered the issue of genetic testing prior to participating in the study. These individuals were given a hypothetical scenario about a genetic form of bone cancer that was found in their uncle, and they were asked to decide whether or not they would want to approach their general practitioner for genetic testing. The decision making process was examined by tracking what information participants viewed through an online program detailing testing information, and by examining their verbal explanations of their thought processes as they were making the decision. Several distinct processes of decision making were identified including: the extent of consideration of alternatives, and the extent of evaluation of the information and

the perceived consequences of the available options. In this study, the most common features that participants viewed during the decision making process were information about genetic test attributes, disease characteristics and treatment attributes.

Furthermore, the main factors that participants said they considered in their decision making process were disease treatment, concerns for family members, and reliability of the test, although other factors were also important for some individuals (Henderson, Maguire, Gray, & Morrison, 2006).

The testing option chosen is important because the test results, or lack of test results, may have downstream medical and psychosocial consequences for the patient post-decision. For example, one study of 465 women who underwent *BRCAl/2* testing found that mutation carriers were significantly more likely to undergo risk-reducing mastectomy, and/or risk-reducing oophorectomy than women who received other test results. The researchers in this study found that more than 80% of carriers had undergone at least one of these risk-reducing surgeries indicating that testing likely has an indirect effect on cancer outcomes (Schwartz et al., 2012). Increased screening behaviors have also been observed in carriers of these mutations (Heshka, Palleschi, Howley, Wilson, & Wells, 2008). In thinking about psychosocial outcomes, one meta-analytic review of post-testing emotional distress in *BRCAl/2* mutation carriers found that carriers experienced increased distress after receiving their results, however, the distress levels returned to pre-testing levels over time (Hamilton, Lobel, & Moyer, 2009). This study also found that distress among non-carriers and those with inconclusive results decreased over time.

Choice Structure

Advancements in decision-making science have occurred in multiple disciplines outside of the medical context including the fields of marketing, economics, and psychology. There are two fundamental paradigms for thinking about decision making evident in the literature. The first of these approaches is called the normative approach which assumes a rational decision maker with a well-defined preference (Mather, Verstraten, & Anstis, 1998). Essentially, this approach assumes that people use the information available to make the decision that is best for them and that the decision would remain consistent every time. The alternative paradigm is called the descriptive approach to decision making and involves examining decisions in practice. Studies utilizing the descriptive approach to decision making have found that peoples' decisions deviate from the rational choice in predictable ways across many scenarios (Mather et al., 1998). This has relevance for providers who facilitate patient decisions because the way they frame and present choices can influence the decision made and its consequences. (Thaler & Sunstein, 2009).

Presentation of Options

One finding with relevance to scenarios in which patients are forced to choose from multiple medically appropriate options is that the number of options presented influences the choice made (Tversky & Kahneman, 1974). In the theory of rational choice, it is implied that a preference between options cannot be altered by the addition of new alternatives. However, there is evidence to suggest that people's preference between two options can depend on the presence or absence of a third alternative (Simonson, 1989). This is thought to occur because the relative attractiveness of options can change as people evaluate them. For example, when there are multiple attractive options, each of

which has advantages and disadvantages, people may delay making a decision, seek to maintain the status quo, or seek additional information. In contrast, adding an additional option which appears less attractive can enhance the attractiveness of the comparison option and increase the likelihood that it is chosen (Mather et al., 1998).

Furthermore, people may evaluate options differently when they are presented simultaneously than when they are presented separately from each other. Changing the evaluation of options from a simultaneous to a separate evaluation can lead to a reversal of preferences. For example, in one study participants were asked to evaluate the appropriateness of a settlement from the sale of a lot owned by two neighbors. Many participants preferred different options when asked to pick the better of the two settlements than when they were presented with each settlement separately and asked to indicate the acceptability of each on a rating scale. For example, 75% of participants judged option A as preferable to option B when evaluating both options simultaneously, and 71% rated option B as better than A when asked to rank each one separately (Bazerman, Loewenstein, & White, 2013). It is hypothesized that some option attributes are easier to evaluate than others and difficult-to-evaluate attributes have a greater impact on joint evaluation than on simultaneous evaluation than easy-to-evaluate attributes (Hsee, Loewenstein, Blount, & Bazerman, 1999).

Giving these findings, it is possible that offering additional genetic testing options, and altering the way these options are presented may influence whether an individual undergoes genetic testing and which test is chosen. However, there has been no work to examine how offering multiple genetic tests for one category of disease, such as hereditary cancer can influence these outcomes. Additionally, it is unclear how people

view the relative attractiveness of genetic panel tests that have tradeoffs correlated with the number of genes tested. For example, testing more genes simultaneously has a greater cost effectiveness and higher likelihood of identifying an underlying genetic predisposition for cancer but there is also a higher likelihood of receiving a result that is uncertain or has unclear clinical utility (Hall et al., 2014). It is also unclear if the same attributes that may drive the choice of one test over another are the attributes most likely to influence medical and psychological outcomes.

Provider Recommendations in Genetic Services

Historically, there has been a paradigm of non-directiveness in genetic services which is different from other medical fields which operated under a model of provider dictated medical care (Elwyn et al., 2001). Under the philosophy of non-directiveness, geneticists and genetic counselors should help clients arrive at decisions based on the client's values and beliefs and not the opinion of the clinician (Elwyn, Gray, & Clarke, 2000). Non-directiveness is in part a reaction to the practice of eugenics and is one way that the field of genetics distinguishes itself from eugenic practices (Elwyn et al., 2000). However, genetics and some other medical fields have been moving away from non-directiveness toward a shared decision-making process between patient and provider (Elwyn et al., 2001). As a part of this process, it may be appropriate for providers to give recommendations about genetic testing for inherited cancer predispositions.

Furthermore, some studies have shown that many patients would prefer a recommendation about genetic testing. One qualitative study about breast cancer susceptibility testing found that some women prefer to have a recommendation and that they differed in whether they preferred to be directed toward a single recommended

course of action, or whether they still wanted to hear all of the options available before making a decision (Geller, Strauss, Bernhardt, & Holtzman, 1997). A survey of 426 women considered at risk for breast cancer, reported that 82% of participants would want providers to make a recommendation about breast cancer susceptibility testing (Geller & Bernhardt, 1998). Another group conducted qualitative interviews of Hispanic women at risk for HBOC and found that different sub-ethnicities differed in how they preferred their provider to make a genetic testing recommendation. For example, groups of Mexican, Cuban and Puerto Rican women differed in whether they would rather have their provider urge them to get a genetic test, ask them to consider testing, or framed their recommendation in terms of what they recommend similar women to do (Vadaparampil et al., 2010).

Patients may not only prefer a provider recommendation, but these recommendations can also be an important factor when patients make testing decisions. For example, in a study of 446 individuals with a known familial HNPCC mutation, 47% of the 299 individuals who underwent testing indicated that a physician's recommendation was important in their decision (Aktan-Collan et al., 2000). Additionally, participants in a population-based study by Bosompra et al., (2000) indicated that participants were more likely to undergo testing with a provider recommendation.

These investigations only account for recommendations of whether or not to undergo genetic testing, they do not examine the influence of provider recommendations in the context of more complex decisions related to which test is chosen.

Hypothetical Vignettes in Genetics Research

There have been numerous studies of patient-related factors that are predictors of testing uptake, mostly in the context of HBOC and Lynch Syndrome in both high risk and general populations. In all of these studies, the decision was either a true or hypothetical binary decision of whether or not to undergo testing. A meta-analysis of 40 studies of genetic testing uptake found that overall, hypothetical uptake was slightly higher (66%) than real uptake (59%).

Hypothetical vignettes are commonly used for studying genetic testing decisions. This methodology has many advantages including cost-effectiveness, speed, and the ability to convey scenarios in a standardized way (Persky, Kaphingst, Condit, & McBride, 2007). However, on a study by study basis, there is often a disparity between predicted and actual genetic testing rates (Persky et al., 2007). Based on a review of 38 articles that used a hypothetical vignette methodology, the authors identified a number of vignette elements related to reliably predicting genetic test uptake accuracy, including strategies to increase the realism of a scenario. Some of these strategies included mentioning a test administrator and making the test seem more temporally imminent. Additionally, they found that the presence or absence of a heredity description significantly influenced interest in testing. Finally, they found that the number of words, sentences, and multisyllabic words were not associated with estimates of testing uptake indicating that they neither improve the accuracy of uptake estimates or undermine the ability to engage with scenarios (Persky et al., 2007).

METHODS

Study Sample

Study Population and Randomization

Potential study participants were recruited through their involvement in the healthy volunteers database at the National Institutes of Health. This database contains individuals who are willing to be contacted about opportunities to participate as healthy volunteers in biomedical research studies through the NIH intramural research program. There were three specifications given to the healthy volunteers office for generating the list from the database. These were that the list should contain approximately 3,000 participants, participants must have text input in the email address field in their database entry, and should reflect a census representative distribution of ages as closely as possible which would be 1710 people in the 18-44 years old group, 870 people in the 45-64 years old group, and 420 in the 65 and over group. These three age strata were determined based on pre-existing classification of age in the database.

The researchers received three lists from the office of healthy volunteers, one for each age strata with 3,083 people on the 18-44 list, 943 on the 45-64 list and 325 on the 65+ list for a combined total of 4,351 potential participants. Participants who had text in the email field that was not an email address (e.g. "NO EMAIL") and duplicate emails were removed from the list.

The researchers then selected some participants from the list provided by the healthy volunteers office to create a census representative age distribution from each age group to create the desired list of approximately 3,000 participants. This involved including all people in the 65+ age strata, because the number of people in this age group was low and a subset of people chosen using a random number generator from the other two age groups. Participants on this list were then assigned to either group 1 or group 2 using random number generation.

A sample size of 400, anticipating a 25% response rate based on previous literature on unsolicited online surveys, was determined to be sufficient to address the study questions. As it was unclear how many emails from the dataset would be viable and what the response rate would be, the investigator decided to invite a subset of people assigned each group to take the survey with the option to invite more people as needed to achieve the sample size target. For this subset, older age groups were oversampled in anticipation of a higher response rate in younger participants due to differences in internet usage patterns. This subset was selected to contain 40% of people in 18-44 age group from the list of 3,000, 60% in the 45-64 age group, and all participants in the over 65 age group which created a list of 1573 total invitees for both groups combined. An additional 7 individuals were randomized to receive an invitation to take either one of the surveys after contacting the researcher requesting participation. The demographics of this sample are reflected in Table 1.

Survey Distribution

The lists of participant emails from each experimental group were uploaded into Qualtrics as panels. An email message was then distributed to each panel. Consequently, each potential participant received an email with a message inviting him/her to participate in the study and a personalized link to the survey that enabled one-time completion of the survey and enabled the participant to return to a partially completed survey (Appendix A). The survey was launched August 29th, 2014 and closed September 14th, 2014 and therefore was active for a total of 17 days. The survey was closed as the number of participants taking the survey had stagnated at zero and the desired sample size had been achieved. Survey distribution statistics are represented in Table 2.

Study Design

The current study uses a randomized trial administered via an online questionnaire in which two experimental groups were presented with a hypothetical genetic testing vignette followed by a series of questions. Provider recommendations were incorporated into the questionnaires and tailored to the participant using survey logic based on an information preference that each participant indicated in one survey question. A flow chart of the study design is found in Figure 2.

Experimental Groups

There were two questionnaire versions. Both questionnaire versions contained the same hypothetical vignette but they differed by the associated method of presenting the genetic testing choice. The same scales and demographic questions were administered to both groups.

Group 1 received survey version 1. Participants in this group were presented with a tiered approach to genetic test presentation. All participants in group 1 were first offered a choice between two options- No Genetic Testing and a 5 Gene Test. After this decision, all participants who indicated that they would want the 5 Gene Test were asked to select an information preference. This involved choosing whether they preferred (a) having only genetic information with clear meaning for their future health care, or (b) having all possible genetic information. After answering this question, participants were presented with a third genetic test- a 15 Gene Test- and a recommendation for either the 5 or 15 Gene Test based on their indicated information preference. They were then given the option to stay with or change their initial genetic test choice. The information preference was elicited after the initial genetic testing choice because this enabled the

study of the impact of the number of options independently from the impact of a personalized recommendation. Participants who had indicated that they did not want genetic testing when presented with the initial genetic testing choice were presented with an alternate question. They were told about the 15 Gene Test and asked if they would have wanted testing if they had been presented with this option.

Participants in group 2 were presented with all three genetic testing options simultaneously - No Genetic Testing, the 5 Gene Test, and the 15 Gene Test. After making the initial genetic testing decision, all participants who indicated that they wanted either the 5 Gene Test or the 15 Gene Test were asked to indicate their information preference using the same question that was presented to group 1. They were then given a personalized recommendation for either the 5 Gene Test or 15 Gene Test based on their information preference and the option to stay with or change which testing option they selected.

The 5 Gene Test and 15 Gene Test varied by the number of genes tested and the following factors: the likelihood of receiving a result that is uncertain, the likelihood of receiving a result with unclear implications for cancer risk, and a different time to test result. The value of each of these attributes increases with more genes tested. These trade-offs between each test would be equivalent to the tradeoffs experienced between undergoing a cancer-specific high penetrance gene panel and a cancer-specific gene panel with high and moderate penetrance genes.

Survey Instrument

The survey instrument was an online questionnaire which contained four sections; informed consent, hypothetical vignette/genetic testing information, genetic testing

preference questions, and scales/demographic questions. The survey was created and administered using Qualtrics Research Suite Software. Screen shots containing the survey content and appearance are found in Appendices B through F.

Initial feedback on the survey was provided by members of the Johns Hopkins/NHGRI Genetic Counseling Training Program executive committee, and one practicing cancer genetic counselor. Revisions were then made and the survey was pilot tested with a convenience sample of 5 community members with limited knowledge of genetics. Changes were made based on their feedback. Based on information from this pilot it was projected that the survey would take 10-15 minutes to complete. The final version of the survey was reviewed by two members of the author's thesis committee prior to launch.

Survey Flow

Participants could not move on to the next question if the initial genetic testing decision was not answered. All other questions were optional; however, participants received a pop up message informing them when they skipped questions with a choice to return and answer those questions or proceed to the next page. Once a participant proceeded to the next page he/she could not return to the previous page. Some questions had randomization of response options.

Consent

Clicking on the survey link directed participants to the first page of the survey instrument which was a consent statement outlining the purpose of the study, survey content, potential risks and benefits, confidentiality information, and the contact

information of the researchers. In order to consent to participate in the study, participants needed to continue to the next page of the survey.

Hypothetical Vignette

A hypothetical vignette was generated based on the author's clinical experience, data from the literature, and quantitative data about the design of hypothetical vignettes for genetic testing. It contained four sections; the hypothetical situation, what could be learned from genetic testing, what would happen if a positive result was found, and the logistics of genetic testing. The hypothetical situation involved the participant receiving a referral to genetic counseling based on a family history that involved early onset breast and prostate cancers in the participant's mother, uncle, and grandfather. The scenario was loosely based on the constellation of cancers seen in HBOC with an overemphasis on prostate cancer to enhance relevance to male participants. In addition to their involvement in HBOC, breast and prostate cancers were chosen because these cancers are common and more likely to be familiar to members of the general population.

Genetic Testing Decisions

Participants were asked to make a series of genetic testing-related decisions as outlined in the study design section of the methods.

Scales

Four pre-existing scales were used to measure the concepts of decisional conflict, intolerance for uncertainty, genetic literacy, and subjective numeracy. Additional questions were used to discern understanding of the scenario, anticipated decisional regret, and genetic comprehension.

Decisional Conflict was measured using a compilation of questions from the Decisional Conflict Scale by AM O'Connor (O'Connor, 1995), and a modified version of this scale tailored to HBOC genetic testing decisions (Katapodi, Munro, Pierce, & Williams, 2011). Neither scale was used in its full form because not all questions made sense in the context of genetic testing and/or a hypothetical scenario. This concept was measured in order to determine if using a tiered versus menu approach to genetic testing related to decisional conflict scores. Decisional conflict was of interest because this is one type of decision making outcome that extends beyond the actual choice made. Anticipated decisional regret was assessed using one question appended to this scale and was added as an additional outcome of decision making and factor of descriptive interest.

Intolerance for uncertainty was measured using the Short Form of the Intolerance for Uncertainty Scale (Carleton, Norton, & Asmundson, 2007). This concept was measured because it is a person-related factor that could potentially explain some variation in outcomes since the genetic tests presented to participants varied in the probability of results with unclear clinical implications.

Genetic familiarity was measured using a modified version of the Rapid Estimate of Adult Literacy in Genetics (REAL-G) for survey form (Erby & Roter, 2008). Genetic comprehension was also measured using questions created from the words on this scale. These scores were administered to determine estimates of the genetic literacy of the study participants and whether genetic literacy and comprehension related to variation in outcomes.

Numeracy was measured using the Subjective Numeracy Scale (Zikmund-Fisher, Smith, Ubel, & Fagerlin, 2007). This concept was measured to gain a sense of the overall

subjective numeracy level of the study participants because the genetic testing decision involved processing numerical and probabilistic information.

A question was added to assess self-reported understanding of the hypothetical vignette and genetic testing scenario. The purpose of this question was to aid assessment of the quality of the vignette based on whether participants felt that they were able to understand it.

Demographics

A series of questions were asked about the participants' experiences with cancer including his/her number of relatives with cancer, perceived cancer risk, anxiety about developing cancer, cancer screening behaviors, and personal and/or family history of cancer genetic testing. Participants were also asked to rate their overall health. These factors were measured because it was hypothesized that a participants' personal experiences with and perceptions of cancer may relate to their genetic testing choices and information preferences.

Additional demographic information was collected including the participants' sex, age, education, income, number of biological children, employment status, race, ethnicity, state of residence, political beliefs, relationship status, and whether his/her native language was English. These were measured as additional person-related factors that could potentially relate to study outcomes and as factors of descriptive interest.

Compensation

Participants received a \$10.00 amazon.com electronic gift card if they entered an email address on the final question of the survey.

Data Analysis

Data Analyses were performed using Stata12 IC. The likelihood of undergoing genetic testing by survey group and the likelihood of a test choice matching information preferences were investigated using bivariate and multivariate logistic regressions. Covariates were included in the multivariate models if they reached statistical significance of $p \leq 0.05$ when a separate regression was run with each variable included as a single covariate. Covariates significant for one genetic testing outcome (i.e. final testing option chosen) were included in the other multivariate analyses to facilitate cross comparisons of the results. The variables sex and age were included in the multivariate analyses for theoretical reasons despite not reaching the inclusion cutoff.

The final genetic test chosen by group was investigated using ordinal logistic regression. The changes in genetic testing choices before and after a provider recommendation were examined using a Wilcoxon matched-pairs signed-rank test using only the data from Group 2.

Variables

Independent variables:

- 1.) Group 1 or Group 2 membership which is equivalent to:
 - a. # of options presented (2 or 3)
 - b. Genetic test presentation method (Step-wise or side-by-side)
- 2.) Genetic testing option chosen- initial choice for Group 2 only
 - a. Answer to “Genetic Testing Choice 1” for Group 2 only (No Testing, 5-Gene Test, 15-Gene Test)
- 3.) Concordance between Information Preference and Initial Genetic Testing Option Selected, Group 2 Only

- a. Match = Selecting:
 - i. 5 Gene Test and (a) having only genetic information with clear meaning for their future health care
 - ii. or 15 Gene Test (b). having all possible genetic information
- b. Mismatch= Selecting the 5 Gene Test and (b) or the 15 Gene Test and (a).

Dependent variables:

- 1.) Decision to have genetic testing or not- initial choice
 - a. Answer to “Genetic Testing Choice 1” made binary (No Testing, Yes Testing)
- 2.) Decision to have genetic testing or not- initial choice accounting for answers of group 1 non-testers when asked if they would want the 15-gene test
 - a. Answer to “Genetic Testing Choice 1” + testing preference for 15 gene test among Group 1 no-testers (No Testing, Yes Testing)
- 3.) Genetic testing option chosen- final choice
 - a. Genetic Test Choice from “Provider Recommendation” + Non Testers from “Genetic Testing Choice” (No Testing, 5-Gene Test, 15-Gene Test)
- 4.) Genetic testing option chosen- final choice , Group 2 only
 - a. Genetic Test Choice from “Provider Recommendation” + Non Testers from “Genetic Testing Choice” for Group 2 only (No Testing, 5-Gene Test, 15-Gene Test)
- 5.) Concordance between Information Preference and Final Genetic Testing Option Selected, Group 2 Only
 - a. Match = Selecting:

- i. 5 Gene Test and (a) having only genetic information with clear meaning for their future health care
 - ii. or 15 Gene Test and (b). having all possible genetic information
 - b. Mismatch= Selecting the 5 Gene Test and (b) or the 15 Gene Test and (a)
 - c. Other= Selecting “No Genetic Testing”
- 6.) Concordance between Information Preference and Final Genetic Testing Option Selected
- a. See explanation under 5.) above for details
- 7.) Decisional Conflict Scale Score
- a. Numerical score calculated from questions on decisional conflict scale

Other demographic variables may be incorporated into multivariate models

TABLES AND FIGURES

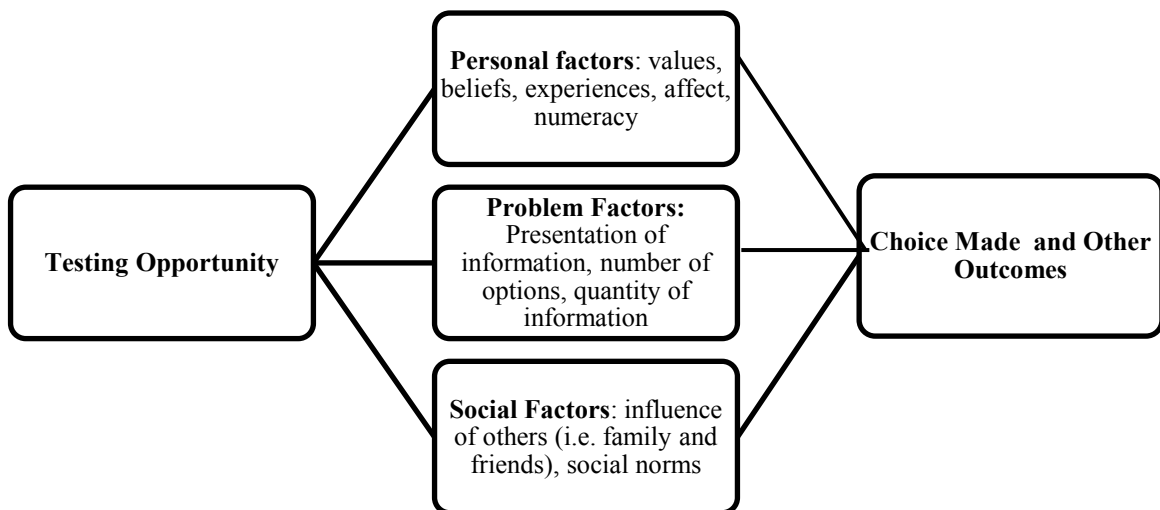


Figure 1. Conceptual framework of factors affecting decision making

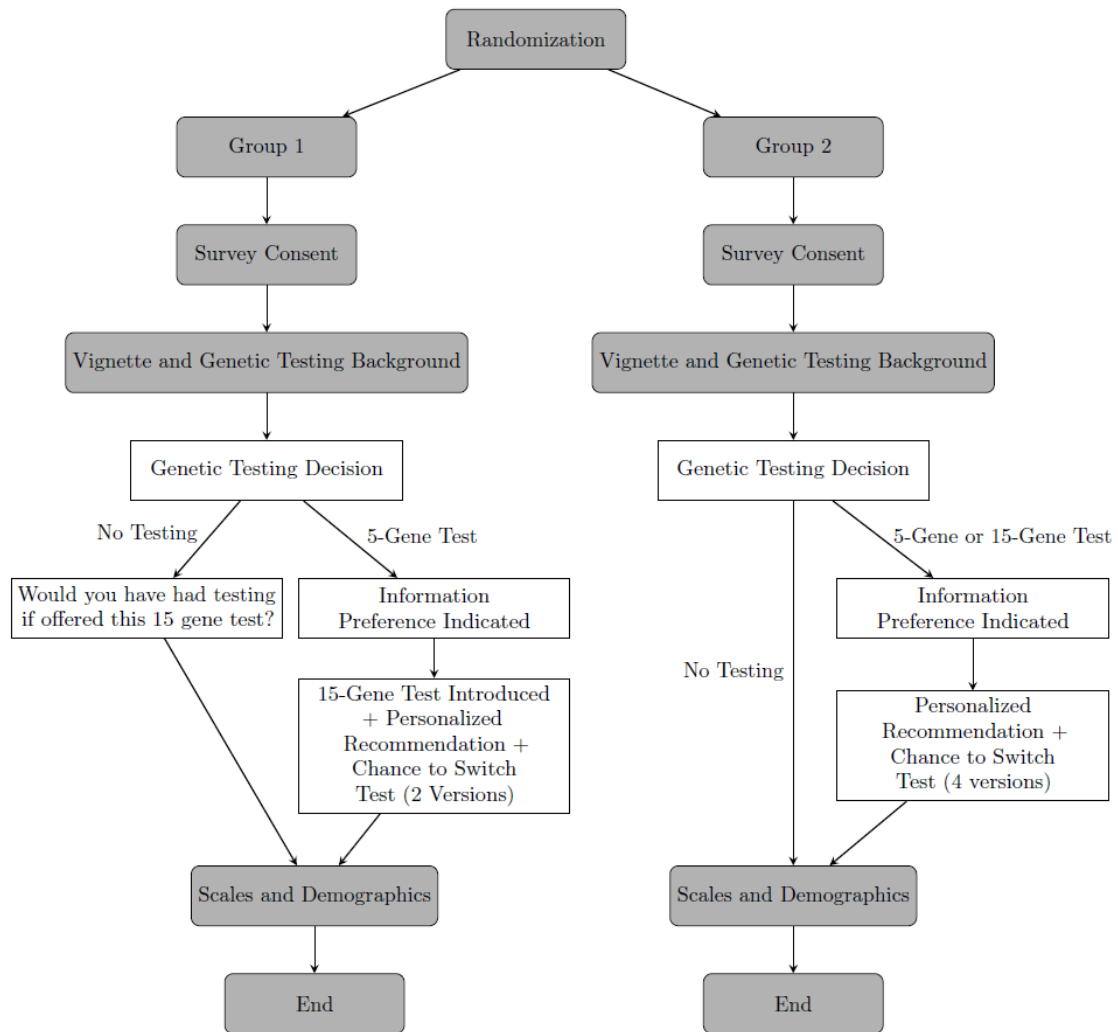


Figure 1. Flow chart of study design. White boxes indicate that the survey question content varied by group. Survey logic was used to administer personalized follow-up questions to the initial genetic testing decision.

		Group 1 - Number of People (%)	Group 2 - Number of People (%)
Age	18-44	360 (45.8)	360 (45.3)
	45-64	271 (34.5)	260 (32.7)
	65+	152 (19.3)	170 (21.4)
	Unknown	3 (0.4)	4 (0.5)
	Total	786	794
Sex	M	340 (43.3)	356 (44.8)
	F	436 (55.5)	423 (53.3)
	Unknown	10 (1.3)	15 (1.9)
	Total	786	794
Race	White	348 (44.3)	389 (50.0)
	Black/AA	301 (38.3)	286 (36.0)
	Asian/Pacific Islander	40 (5.1)	37 (4.7)
	Hispanic	49 (6.2)	39 (4.9)
	Native American	5 (0.6)	3 (0.4)
	Multiple Races	17 (2.2)	16 (2.0)
	Unknown	26 (3.3)	24 (3.0)
	Total	786	794

Table 1. Demographics of sample invited to take the survey.

	Randomization	
	Group 1 N, (% of Initial Dataset)	Group 2 N, (% of Initial Dataset)
Initial Dataset	786, (100)	794, (100)
Email Distribution List after Loading into Qualtrics	780, (99.2)	790, (99.5)
Emails Bounced	102, (13.0)	89, (11.2)
Opened Email	425, (54.1)	425, (53.5)
Clicked on Survey Link	263, (33.5)	250, (31.5)
Consented to Study	248, (31.6)	237, (29.8)
Completed Survey	215, (27.4)	204, (25.7)

Table 2. Survey distribution statistics by group

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PART 2: MANUSCRIPT

ABSTRACT

Objective:

To examine how the presentation of a decision can influence choices about genetic testing for inherited cancer predispositions. Specifically, how the number of options and the addition of a personalized recommendation might influence outcomes such as the likelihood of undergoing genetic testing, the genetic test chosen, and whether a person's test choice matches their personal preferences.

Methods:

An online hypothetical vignette study was completed by 454 healthy volunteers. Each participant was randomized to receive one of two survey versions which differed in the manner of presenting testing options and how these options were integrated with a provider recommendation. Regression analyses were performed to determine the relationships between the presentation of choice and participant decisions. Wilcoxon rank-sign tests were used to determine the impact of a provider recommendation on final genetic testing choices.

Results:

Participants were more likely to choose to have genetic testing when presented with three options instead of two (OR: 2.00 $p=0.014$). This effect was no longer observed when individuals who had decided not to undergo testing were presented with a third option (OR: 0.90 $p=0.775$). The addition of a provider recommendation did not significantly change the overall distribution of options chosen ($p=0.746$). However, after a

recommendation, participants were more likely to choose the test that best matched with personal preferences about the type of genetic information desired ($p < 0.001$).

Conclusions:

Participants were more likely to undergo genetic testing when presented with 3 options instead of 2. They were also more likely to select an option in line with a personal preference if presented with a recommendation based on this preference.

Practice Implications:

The way providers' structure preference-based medical choices for patients and the advice they give may influence which options are selected.

1.0 INTRODUCTION

Genetics is one area of medicine in which many new medical testing options have become available in a short period of time due to rapid technological advancements. The abundance of clinical testing options has made decision making about genetic testing more complex for patients and providers. Furthermore, the way providers choose to structure and present genetic testing choices may influence the ultimate decisions that clients make and their subsequent medical care. The objective of the following hypothetical vignette study is to examine how the structure and presentation of genetic testing, specifically the number of testing options and a personalized recommendation from a health care provider, can influence the genetic test chosen in regard to hereditary cancer predispositions.

1.1 Inherited Cancers

Some individuals are born with a genetic mutation that places them at an increased lifetime risk to develop specific types of cancer, often at younger ages than

people in the general population. These individuals are said to have a hereditary cancer syndrome, and it is believed that about 5-10% of all cancers are inherited via this mechanism (Garber & Offit, 2005). There are more than 45 identified syndromes with clear genetic causes that confer an increased lifetime risk of developing cancer (Riley et al., 2012). These syndromes can differ on many dimensions including the types of cancer, the magnitude of the risks, the inheritance pattern, and whether individuals have other physical symptoms.

One of the most common hereditary cancer syndromes is called Hereditary Breast and Ovarian Cancer Syndrome (HBOC). HBOC is caused by a deleterious mutation in either the *BRCA1* or *BRCA2* gene (Garber & Offit, 2005). This syndrome is associated with elevated lifetime risks of developing certain cancers including up to an 80% lifetime chance to develop female breast cancer, 40% chance to develop ovarian cancer, and elevated risks for prostate and pancreatic cancers (Petrucelli, Daly & Feldman, 2013). Approximately 1 in 800 women in the general population and 1 in 40 women of Ashkenazi Jewish ancestry have this condition (Schneider, 2012).

1.2 Genetic Testing for Inherited Cancers

Genetic testing can be used to identify individuals with hereditary cancer syndromes and inform their cancer risk assessments. Typically, a person would come to clinical attention as a candidate for hereditary cancer testing based on a notable family or personal history of cancer or if a mutation is identified in a relative. Individuals identified as having elevated risks for cancer by genetic testing may use these results to guide medical management decisions about cancer treatment, screening and/or prevention with the objective of reducing cancer morbidity and mortality. For example, women identified

as having HBOC may decide to have increased breast surveillance or have prophylactic surgeries such as a bilateral mastectomy or salpingo-oophorectomy due to their high risk of breast and ovarian cancers. Genetic test results can also be useful for guiding genetic testing in relatives and informing their cancer risks.

Although some information learned from genetic testing has clinical utility, genetic testing can also provide information with unclear implications for clinical care. For example, it is possible to learn about risks for cancers that cannot be screened for or prevented. The interpretation of genetic testing results is also limited by incomplete supporting information. Testing of genes that have not been rigorously studied and/or confer moderately increased risks for cancer can lead to a result that does not have clear implications for cancer risks, surveillance and/or prophylactic surgeries. Additionally, testing of any gene can return a result of uncertain clinical significance. This means that an individual has a genetic change that has not been observed before or is not sufficiently understood to be classified as disease causing or not. Furthermore, a negative genetic test result does not indicate a decreased risk of cancer unless a family member has previously been found to have a deleterious mutation in the gene examined.

Genetic testing for inherited cancers has become more complex over time as next-generation sequencing technologies have enabled companies to offer various panel tests that contain multiple genes (Hall et al., 2014). At this time there are four general approaches to cancer genetic testing including the following:

- 1.) Syndrome specific test (e.g. testing for HBOC)
- 2.) Cancer-specific high penetrance gene panel (e.g. genes for several syndromes that cause a high-risk for breast cancer)

- 4.) Cancer-specific gene panel with high and moderate penetrance genes (e.g. many genes that have some association with increased breast cancer risk)
- 5.) “Comprehensive” cancer panels that include genes associated with multiple cancers or hereditary cancer syndromes (e.g. genes associated with HBOC, Lynch Syndrome and other cancer syndromes) (Hall et al., 2014).

Each of these approaches has distinct clinically relevant advantages and disadvantages which constitute trade-offs in choosing one method over another. Generally, it is up to patients to decide if they would like to have genetic testing, and in cases with multiple medically appropriate tests, which test is the best fit for their preferences (Riley et al., 2012). This process can be challenging for both patients and providers.

1.3 Decision Making

Studies of decision making have found that people are not completely rational decision makers and their decisions deviate from rational choice in predictable ways across many scenarios (Mather et al., 1998). This has relevance for how providers present information upon which a medical decision will be based (Thaler & Sunstein, 2009). In the case of genetic testing, the way health care providers choose to structure and present testing choices to clients may influence the choice made.

Findings suggest that the number of options presented to people when making a decision, for instance when patients are forced to choose from multiple medically appropriate options, affects the decision made (Tversky & Kahneman, 1974). In the theory of rational choice, it is implied that a preference between options cannot be altered by the addition of new alternatives. However, there is evidence to suggest that people’s

preference between two options may depend on the presence or absence of a third alternatives (Simonson, 1989). This is thought to occur because the relative attractiveness of options changes the way people evaluate them.

Another way in which providers can influence a patient's medical decision is whether or not they provide a recommendation. A population-based study by Bosompra et al., (2000) indicated that participants were more likely to have genetic testing with a provider recommendation. Furthermore, a survey of 426 women considered at risk for breast cancer, reported that 82% of participants would want providers to make a recommendation about breast cancer susceptibility testing (Geller & Bernhardt, 1998). Patients may also differ in the type of recommendations they prefer. For example, a qualitative study about breast cancer susceptibility testing found that among those who desired a provider recommendation, preferences varied between hearing a single recommended course of action or all of the options available before making a decision (Geller et al., 1997).

This study seeks to investigate whether the ways in which genetic testing choice is presented can influence which tests are chosen and the likelihood of undergoing genetic testing in the context of hypothetical hereditary cancer predisposition testing. Specifically, this study was designed to examine if the number of testing options offered and an accompanying provider recommendation for one of the test options would influence a hypothetical decision about genetic testing for hereditary cancer predisposition.

2.0 METHODS

2.1 Study Population and Randomization

A list of potential study participants was obtained from the healthy volunteers database at the National Institutes of Health. This database contains individuals who are willing to be contacted about opportunities to participate as healthy volunteers in biomedical research studies through the NIH intramural research program. A subset of people from this list were selected for email invitation using random number generation with oversampling of older age groups (relative to a census representative distribution) to account for anticipated differences in internet usage patterns. Potential participants were randomized to an experimental group (Group 1 or Group 2) using random number generation. Exclusion criteria included being under 18 years of age and not having an email address in the database.

2.2 Survey Distribution

Potential participants received an email invitation to participate in the study with a personalized link to the online survey. Each link enabled one-time completion of the survey and allowed the participant to return to a partially completed survey. The survey was active for a total of 17 days in August and September 2014. The survey was closed because the number of participants taking the survey had stagnated at zero over time and the desired sample size had been achieved. Participants had the option to receive a \$10.00 gift card incentive for survey completion.

2.3 Study Design and Survey Instrument

This quantitative study was a randomized trial with two experimental groups. Each group was presented with a hypothetical genetic testing vignette followed by a series of genetic testing decisions, scales and measures, and demographic questions.

Groups differed by the questions that elicited genetic testing decisions, however, the hypothetical vignette and all other survey questions remained constant.

The experimental portion of the survey had two components. The first component was designed to determine the impact of the number of options on the likelihood of undergoing genetic testing. The second component was to investigate the impact of a personalized provider recommendation on genetic testing decisions. A flow chart of the survey design is reflected in Figure 1.

2.3.1 Hypothetical Vignette

A hypothetical vignette was generated based on data about genetic testing for hereditary cancer predisposition in the literature, the author's clinical experience, and quantitative data about the design of hypothetical vignettes for genetic testing (Figure 2). The scenario was loosely based on the constellation of cancers seen in HBOC with an overemphasis on prostate cancer to enhance relevance to male participants. In addition to their involvement in HBOC, breast and prostate cancers were chosen because these cancers are common and more likely to be familiar to members of the general population. Before launch, the survey was reviewed by five individuals without a scientific background.

2.3.2 Genetic Testing Options

There were three possible genetic testing options that were designed to be equivalent to the choice between no testing, a cancer-specific high penetrance gene panel and a cancer-specific gene panel with high and moderate penetrance genes. These options are described in Table 1. The genetic tests differed on three main categories correlated with the number and type of genes including the likelihood of receiving a result that is

uncertain, the likelihood of receiving a result that has unclear implications for cancer risk management, and a different time to test result.

2.3.3 Number of Genetic Testing Options

The first genetic testing decision that participants made was designed to investigate the impact of the number of options presented (2 versus 3) on the likelihood of undergoing genetic testing. Group 1 was presented with two genetic testing options; “No Genetic Testing” or a “5 Gene Test”. Group 2 was presented with a third option of a “15 Gene Test” in addition to the two options presented to Group 1. Participants who chose “No Genetic Testing” in group 1 were asked a follow-up question about whether they would have wanted genetic testing if they had been offered the “15 Gene Test”.

2.3.4 Information Preference and Personalized Provider Recommendation

Following their initial genetic testing decision, all participants who chose to have a genetic test were asked to indicate an information preference for either (a) *only genetic information with clear meaning for future health care* or (b) *having all possible genetic information*. Participants who indicated a preference for “only genetic information with a clear meaning for future health care” received a personalized recommendation for the “5 Gene Test” and participants who preferred “having all possible genetic information” received a recommendation for the “15 Gene Test”. Each participant who received a recommendation was given the option to remain with their initial choice or switch to another option. Participants in group 1 who had opted for genetic testing were introduced to the “15 Gene Test” in conjunction with the personalized recommendation. For participants in group 2, a personalized recommendation was the only intervention between their initial and final genetic testing choices.

2.3.5 Overall Method of Testing Presentation and Final Genetic Testing Choice

The overall method of testing presentation reflects which survey version participants received and therefore how the options and a provider recommendation were presented to participants. The impact of the overall method of testing presentation is illustrated by individuals' final genetic testing choices. The final genetic test choice each participant made was calculated using the final genetic test that each participant chose while taking the survey.

Overall, group 1 participants received a tiered approach to genetic testing presentation and group 2 participants received a menu approach to genetic testing presentation. Group 1 participants were initially presented with two options, and then participants who had chosen the "5 Gene Test" were presented with a third option in conjunction with a provider recommendation and the opportunity to change their choice. Group 2 received a menu approach to genetic testing as they were initially offered all options and testers were later presented with provider recommendation and the option to change or remain with their initial choice.

2.3.6 Measures and Demographics

Decisional conflict was one decision making outcome of interest. It was measured using a compilation of questions from the Decisional Conflict Scale by AM O'Connor (O'Connor, 1995), and a modified version of this scale tailored to HBOC genetic testing decisions (Katapodi et al., 2011). Neither scale was used in its full form because only subsets of questions made sense in the context of genetic testing and/or a hypothetical scenario.

Three additional scales were used to measure the concepts of intolerance for uncertainty, genetic literacy, and subjective numeracy. Participants were also asked questions created by the authors to assess genetic comprehension, decisional regret, and self-reported understanding of the hypothetical vignette. These concepts were measured to be used for descriptive analyses and covariates.

Intolerance for uncertainty was measured using the Short Form of the Intolerance for Uncertainty Scale (Carleton et al., 2007). This concept was measured because it is a participant attribute that could have relevance to genetic testing decisions due to the inherent uncertainty associated with the outcomes of a decision and the probabilistic nature of genetic information.

Genetic familiarity was measured using a version of the Rapid Estimate of Adult Literacy in Genetics (REAL-G) (Erby & Roter, 2008) modified for survey form. Genetic comprehension was also measured using questions created using the words from the REAL-G. These scores were measured to determine estimates of the genetic literacy of the study participants.

Numeracy was measured using the Subjective Numeracy Scale (Zikmund-Fisher et al., 2007). This concept was measured to gain a sense of the overall subjective numeracy level of the study participants because the genetic testing decision involved processing numerical and probabilistic information.

A question was added to assess self-reported understanding of the hypothetical vignette and genetic testing scenario. The purpose of this question was to aid the assessment of vignette quality based on whether participants felt that they were able to

understand it. Anticipated decisional regret was assessed using an additional question in the same format as the decisional conflict scale.

A series of further questions were asked about the participants' actual experiences with cancer including his/her number of relatives with cancer, perceived cancer risk, anxiety about developing cancer, cancer screening behaviors, and personal and/or family history of cancer genetic testing. Participants were also asked to rate their overall health. These factors were measured because it was hypothesized that a participants' personal experiences with and perceptions of cancer may relate to their genetic testing choices and information preferences.

Other demographic information was collected including the participants' sex, age, education, income, number of biological children, employment status, race, ethnicity, state of residence, political beliefs, relationship status, and whether his/her native language was English.

2.4 Data Analysis

Data analyses were performed using Stata12 IC. The likelihood of undergoing genetic testing by survey group and the likelihood of a test choice matching information preferences were investigated using bivariate and multivariate logistic regressions. The final genetic test chosen by group was investigated using ordinal logistic regression. Covariates were included in the multivariate models if they reached statistical significance of $p \leq 0.05$ when a separate regression was run with each variable included as a single covariate. Covariates significant for one genetic testing outcome (i.e. final testing option chosen) were included in the other multivariate analyses to facilitate cross comparisons of the results (Table 3). The variables sex and age were included in the

multivariate analyses for theoretical reasons despite not reaching the inclusion cutoff. The changes in genetic testing choices before and after a provider recommendation were examined using a Wilcoxon matched-pairs signed-rank test using only the data from group 2.

3.0 RESULTS

3.1 Sample Description

Of the 1580 emails sent to potential participants 485 (30.7%) individuals consented to the study and 419 (26.5%) completed the survey. The characteristics of participants are shown in Table 2. 93% (N=437) of individuals indicated they agreed or strongly agreed with the statement “I feel like I understood the medical scenario and genetic testing choice”.

3.2 Impact of the Number of Genetic Testing Options

79.4% of individuals in group 1 and 88.7% of individuals in group 2 chose to undergo genetic testing when first asked. Among group 2 participants who decided to undergo testing, 38.0% chose the “5 Gene Test” and 50.7% chose the “15 Gene Test”. Overall, participants who were given three options (group 2) were significantly more likely to undergo genetic testing than participants who were given two options (group 1) (Table 3). Participants who did not opt for testing in group 1 were then asked if they would have wanted genetic testing if presented with an additional option. 48.9% of these individuals said they would have wanted genetic testing if they had been told about the “15 Gene Test”. If these individuals are counted as choosing to undergo genetic testing there is no longer a statistically significant difference in the likelihood of undergoing genetic testing by group (Table 3).

3.3 Impact a Personalized Provider Recommendation

Responses of group 2 participants were used to determine the influence of a provider recommendation on genetic testing choice since this was the only factor manipulated between the initial and final testing decisions in this group. The overall distribution of genetic testing options chosen was not significantly different after a provider recommendation compared to before ($p=0.746$). However, test choice after a provider recommendation was significantly more likely to match the participant's indicated information preference than before ($p<0.001$) (Table 4).

3.4 Impact of the Overall Method of Testing Presentation

There was no statistically significant difference between groups in the likelihood that the final testing option selected matched the participant's information preference (Table 3). Additionally, there was no significant difference in the overall distribution of genetic testing options selected by group. However, members of group 2 were significantly more likely to choose genetic testing than members of group 1 (Table 3). In group 1, 21.5% of individuals opted for "No Test", 29.6% opted for the "5 Gene Test" and 48.9% opted for the "15 Gene Test". Among group 2 members, 11.8% opted for "No Test", 36.2% opted for the "5 Gene Test" and 52.0% opted for the "15 Gene Test" (Figure 4). There was no significant difference in decisional conflict score by group ($P=0.92$).

4.0 DISCUSSION AND CONCLUSION

4.1 Discussion

Participants presented with 3 genetic testing options were more likely to choose genetic testing than participants offered only 2 testing options. This effect was mitigated

by introducing the “15 Gene Test” to individuals who had opted against genetic testing and asking if they would have wanted testing if presented with this option. It is unclear if this discrepancy is due to something uniquely attractive about the 10 additional genes on the “15 Gene Test” or if learning about an additional option changed the way people thought about the decision to have genetic testing or not by adding a point of comparison. These alternative interpretations could be addressed with future studies that add a “15 Gene Test” vs “No Test” control or evaluate the relative attractiveness the attributes that varied between the testing options.

The majority of individuals initially chose a test that was the best match for their information preferences. Specifically, among participants opting for genetic testing, most individuals desiring as much genetic information as possible chose the “15 Gene Test” and those preferring only genetic information with clear meaning for their clinical care chose the “5 Gene Test”. However, a subset of individuals did not initially select a genetic test that matched their information preference. A follow-up of the initial testing decision with a personalized recommendation that advocated for a test selection that matched this preference increased the likelihood that there was a match between the final test chosen and the preferred type of information. This type of recommendation had the largest impact on the subset of individuals who initially selected a test that was not in line with their information preference, but not all individuals with a mismatched test and information preference changed their testing choice in response to the recommendation.

Among those individuals who changed their choice in response to the recommendation, it is possible that the recommendation encouraged them to think more about what they wanted to learn from testing and what they would do with the genetic

information. It is also possible that some individuals did not initially have a strong preference for one test over another and the recommendation tipped the balance in favor of one test causing them to change their selection. Forcing participants to reconsider their initial choice could also lead to people second guessing their decisions and changing their choices.

Individuals with a discordant information preference and final test choice may have considered another attribute of genetic testing more important than the information obtained from testing. For example, a person may want as much information as possible but strongly desire a test that has a faster turnaround time. In this case, one may decide that the anxiety of waiting several additional weeks for a result is worse than not getting the information from a few additional genes.

Although more individuals overall opted for the “15 Gene Test”, a sizable number still chose the “5 Gene Test”. This indicates that adding additional genes to a test does not ensure that that test will be more desirable and there may be a reason to continue to offer smaller panels of well-studied genes. Additionally, there was no evidence to indicate that offering an additional option which increases the complexity of the decision significantly influences decisional conflict.

4.2 Conclusion

This study indicates that offering a third genetic testing option to all individuals may increase the likelihood that an individual will undergo genetic testing.

Consequently, individuals who receive a tiered approach to testing and do not initially opt for the “5 Gene Test” would not have exposure to the “15 Gene Test” which decreases the overall likelihood of undergoing genetic testing with this approach.

This study also provides evidence that a personalized recommendation targeting information preferences may influence the testing choices of those individuals with a discrepancy between what type of information they prefer to learn and their testing choice. Finally, the overall method of offering testing, a tiered versus a menu approach, did not significantly impact whether a person selected the “5 Gene Test” or the “15 Gene Test”. It also did not significantly influence decisional conflict ratings.

4.3 Limitations

This study has several limitations. One limitation is that the hypothetical nature of the vignette and genetic testing decisions differ from real-life genetic testing contexts. The patient-provider relationship and the social accountability that could stem from interpersonal communication are lost online. Additionally, study participants likely did not have the same affective states of patients in a real-life setting who would be discussing difficult topics such as their family and personal histories of cancer and risks for future cancers. Participants in the study would also not have been anticipating the receipt of actual genetic test results. Additionally, the findings from this may not apply if a decision like this is presented in a very different way, for example, if every syndrome and/or gene contained on a panel test is discussed in detail with patients.

Furthermore, although this study represented people of different ages and races, the individuals in the NIH healthy volunteers database are likely different than other general population samples and are concentrated in the Washington District of Columbia Metropolitan Area.

4.4 Practice Implication

The way that providers present genetic testing choices to clients may influence the choices made. It may be appropriate to present multiple testing options as people have different preferences for the types of information they value. This is particularly relevant in settings with multiple medically appropriate tests with distinct trade-offs. Encouraging patients to talk through how they value these trade-offs and the different downstream implications of potential test results is one approach that can be used to help facilitate informed decision making and give providers the information needed to make personalized recommendations. This study indicates that personalized recommendations using a similar approach may help people who have selected options that are not in line with their preferences. Although this study did not collect information to elucidate the underlying reason for this finding it would be one potential area for further study.

5.0 FIGURES

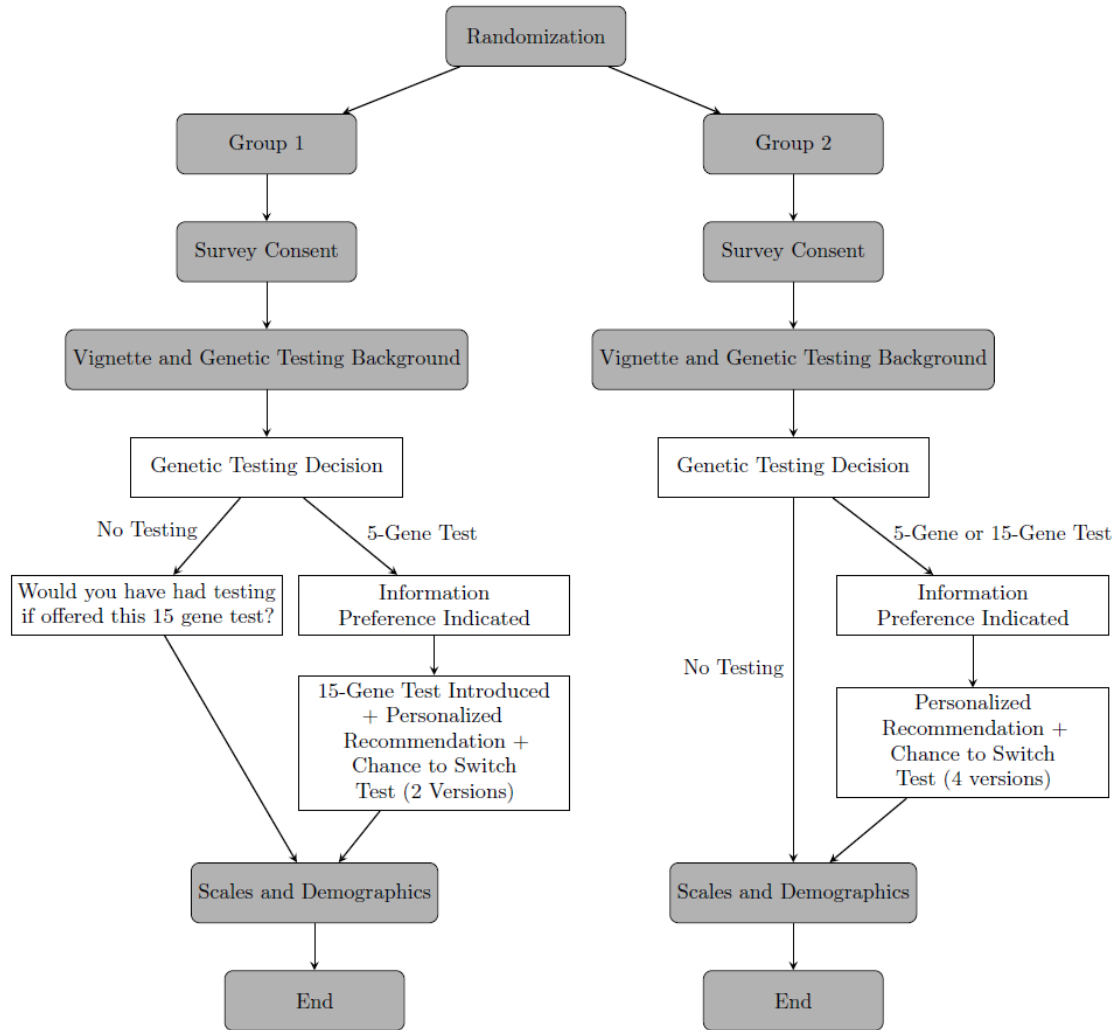


Figure 1. Flow chart of study design. White boxes indicate that the survey question content varied by group. Survey logic was used to administer personalized follow-up questions to the initial genetic testing decision.

This survey is based on a choice that is possible but not actually available to anyone. This survey should not be used as a source of medical fact. You cannot return to previous pages in the survey after hitting ">>" at the end of each page.

Your Situation

Please imagine that you have visited your family doctor to talk about your chance of having or developing cancer. You have told him that your mother, uncle and grandfather all had cancer, either breast or prostate cancer, when they were around 40 years of age. Your doctor is concerned about your family history and suggests that you see a genetic counselor to learn about genetic testing. The purpose of genetic testing is to see if you have a faulty gene that puts you at high risk for certain types of cancer.

When you visit the genetic counselor she asks you questions about your family's cancer history. Based on this, she says that you qualify for genetic testing. She says that not everyone wants genetic testing because the results can be upsetting. It is up to you to decide if you want genetic testing or not. In the next question you will be asked to make a (hypothetical) decision about undergoing genetic testing. Below is some information about genetic testing to help you with your decision.

What You Could Learn from Genetic Testing

You can get three types of results:

<i>Type of Result</i>	<i>Explanation</i>	<i>Meaning for your Cancer Risk</i>
Positive Result	You have a <u>faulty gene</u>	You are at higher risk get certain cancers in your lifetime than people without a faulty gene. For the faulty gene with the biggest cancer risk, about 8 in 10 people will get cancer. Some cancers you could learn you are at risk for are breast, prostate, pancreatic and ovarian cancers.
Negative Result	<u>No faulty gene</u> was found	You may not have inherited a faulty gene that gives you a high risk to have certain cancers. However, your family history may still put you at higher risk than the general public to get certain cancers. You could still have a faulty gene that was not tested.
Inconclusive Result	You have a change in a gene but it is <u>unclear</u> if this increases your risk of cancer.	An unclear result means that the test cannot help determine your cancer risk at this time. Your family history may still put you at higher risk than the general public to get certain cancers. You could still have a faulty gene that was not tested.

What Would Happen if You Have a Positive Result

If you have a positive result (faulty gene found):

- You will be told about your chance to develop certain cancers.
- Doctors may recommend that you have certain tests to look for cancer (such as a mammogram or PSA testing), or surgeries to prevent cancer.
- There may not be ways to find early and prevent every cancer you could be at high risk for.
- Each of your children would have a 50% chance of having the same faulty gene.
- Other family members may also have the same faulty gene and a high cancer risk.

Logistics

- Genetic testing is done on a tube of your blood.
- There is 100 dollar copay for any type of genetic testing.

Figure 2. Hypothetical vignette as it appeared to all study participants.

No Genetic Testing		5 Gene Test	15 Gene Test
	Description	Looks at the 5 best-studied genes that can cause breast and prostate cancers	Looks at the 5 best-studied genes that can cause breast and prostate cancers and 10 other genes that have been tied to breast or prostate cancers
	Quality of information and recommendations if a positive test	Clear guidelines telling your cancer risks and suggestions for screening	Clear guidelines telling your cancer risks and suggestions for screening for 5 genes. Meaning of a positive result for the 10 other genes is not well-established.
	Chance of an inconclusive result	2 in 100 people have an inconclusive result	15 in 100 people have an inconclusive result
	Time it takes to get your results	3 weeks	8 weeks

Table 1. Genetic testing options and descriptions presented to study participants.

Characteristic	Group 1	Group 2
Age - % (N=211, 201)		
- 18-44	46.0	44.3
- 45-64	33.6	29.9
- 65+	20.4	25.9
Sex -% Female (N=214, 204)	58.9	59.8
Race -% (N=212, 203)		
- White	59.0	70.0
- Black or African American	30.2	20.7
- Other	11	9.5
Ethnicity -% Hispanic (N=214, 204)	5.1	3.4
≥1 year of college - % (N=214, 204)	92.1	94.1
Employment - % (N=213, 204)		
- Part-time or full-time	65.7	66.2
- Retired	15.5	19.1
- Unemployed or disabled	14.1	10.8
- Job free by choice	4.7	3.9
Native English Speaker -% (N=214, 204)	86.9	89.7
Relationship Status - %(N=214, 202)		
- Married/Domestic Partnership	44.4	39.1
- Divorced/Separated/Widowed	15.4	19.8
- Single	40.2	41.1
Biological Children -% with (N=213, 204)	50.7	46.1
Subjective Numeracy Score –mean (N=215, 207)	4.76	4.80
% Correct Genetic Comprehension Questions - mean (N=215, 207)	92.2	92.5
Genetic Familiarity - mean score (N=217, 208)	5.9	5.8
Personal History of Cancer - % yes (N=215, 206)	7.9	7.8
One or More First Degree Relatives with Cancer History - % yes (N=215, 206)	45.1	45.1
Personal History of any Cancer Screening - % yes (N=214, 206)	81.3	85.0
Personal or Family History of Cancer Genetic Testing - % yes (N=214, 206)	10.3	9.7
Perceived Cancer Risk Relative to People of Same Age and Gender -% (N=214, 206)		
- Higher	10.7	11.2
- Lower	52.3	52.9
- Same	36.9	35.9
Level of Anxiety about Developing Cancer - % (N=214, 206)		
- High	1.9	4.9
- Moderate	23.4	27.7
- Low	74.8	67.5
Perceived Health - % Good or Excellent (N=214, 206)	93.5	94.7

Table 2. Study participant demographics.

	Bivariate		Multivariate+	
	Odds Ratio	p-value	Odds Ratio	p-value
Initial likelihood of undergoing genetic testing (2 vs 3 options)	2.03	0.008*	2.00	0.014*
Initial likelihood of undergoing genetic testing (2 vs 3 options) accounting for Group 1 non-testers who would want the “15 Gene Test”	0.94	0.843	0.90	0.755
Final likelihood of undergoing genetic testing (tiered vs menu approach)	2.05	0.006*	2.02	0.012*
Final option chosen	1.31	0.135	1.30	0.160
Final option concordance with information preference	0.63	0.069*	0.62	0.080*
*statistically significant ($p \leq 0.05$) +multivariate model controlling for sex, age, race, education, income, employment status, native English speaking, cancer screening behavior, perceived personal cancer risk, genetic comprehension, subjective numeracy, genetic familiarity, intolerance for uncertainty, not completing the survey				

Table 3. Genetic testing decision outcomes by survey group membership.

		Final Test Choice and Information Preference			
		Match	Do not Match	Other	
Initial Test Choice and Information Preference	Match	134	9	1	144
	Do not Match	28	24	0	52
	Total	162	33	1	196

Table 4. Concordance between participant information preference and genetic testing choices before and after a provider recommendation for group 2 participants.

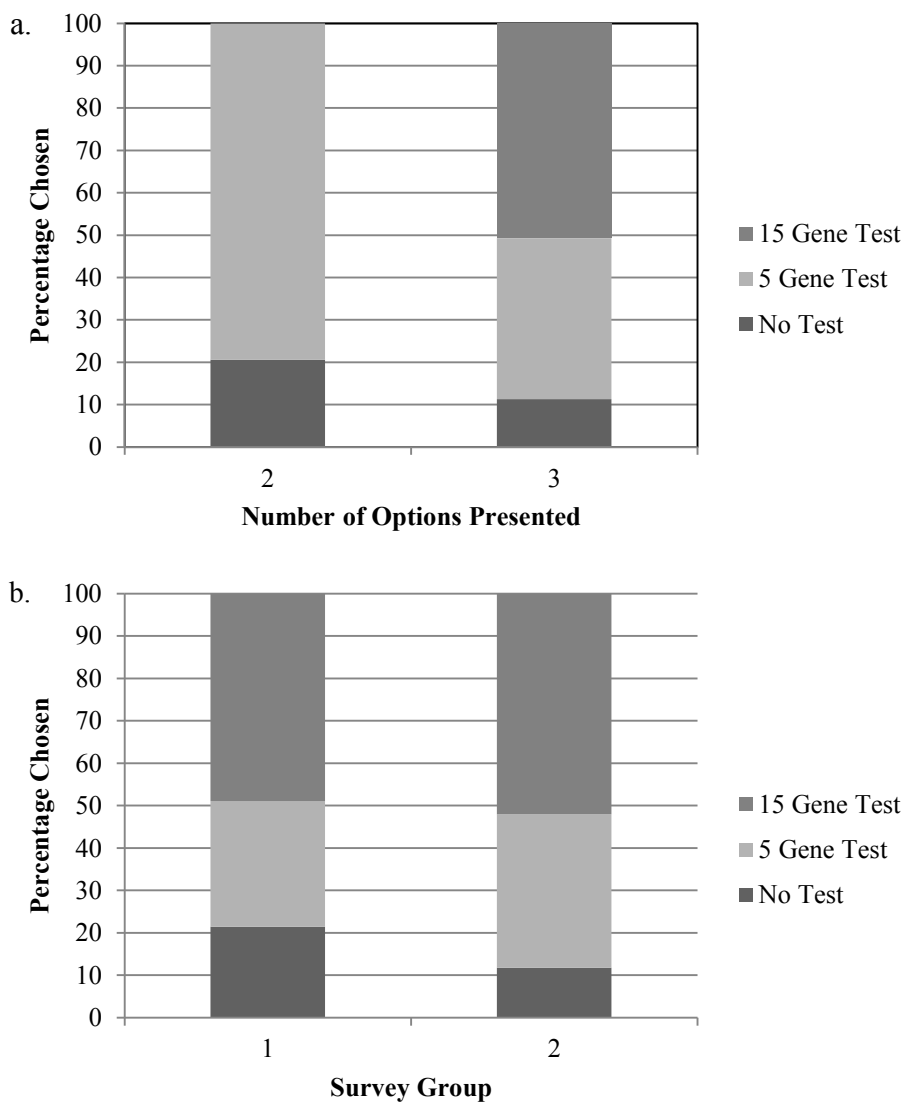


Figure 4. Genetic testing options selected. a. The percentage of each option selected during the initial genetic testing choice by the number of testing options presented. b. The percentage of each genetic testing option selected for the final genetic testing choice by survey group.

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APPENDICES

APPENDIX A: Invitation Email

Hello,

My name is Marci and I am a researcher at the National Institutes of Health in the National Human Genome Research Institute. I received your name from the healthy volunteers program at the NIH and I would like to invite you to participate in study *Decisions about Genetic Testing for Hereditary Cancer Predisposition*. To participate in this study you would need to take a one-time survey. The survey should take 15-25 minutes to complete and you will receive a \$10.00 electronic gift card to Amazon.com if you provide your email at the end of the survey.

To take the survey, please click on the following link (Insert Link Here).

Thanks,

Marci Barr

APPENDIX B: Survey Consent

Informed Consent

Decisions about Genetic Testing for Hereditary Cancer Predisposition

Research Purpose

The purpose of this research is to learn how people make decisions about genetic tests for cancer risks.

Survey Content

You will be asked about...

- what you would decide if you were offered different types of genetic tests
- your personal and family cancer history
- such things as your age, race and income

Risks

It is possible that this survey might make you think about cancer and this might cause worry or concern.

Benefits

You will receive a \$10.00 electronic gift card to Amazon.com if you enter your email at the end of the survey. Otherwise, you will not directly benefit from participation in the study. Findings from this study may help improve care for others in the future.

Confidentiality

Any information about yourself that you provide while taking this survey will only be used for this study. Your email address will be removed from your survey answers before analyzing the data.

Contact

This research has been reviewed by the National Institutes of Health Office of Human Subjects Research. It is being done by researchers at the National Human Genome Research Institute. Please email Marci Barr at barrml@mail.nih.gov with any concerns or comments about this survey.

Participation

If you agree to take part in the survey, click ">>" in the bottom right hand corner. If you do not want to take the survey, please close the survey window in your web browser.

By clicking ">>" you consent to take the survey.

>>

Survey Completion
0% 100%

APPENDIX C: Hypothetical Vignette

This survey is based on a choice that is possible but not actually available to anyone. This survey should not be used as a source of medical fact. You cannot return to previous pages in the survey after hitting ">>" at the end of each page.

Your Situation

Please imagine that you have visited your family doctor to talk about your chance of having or developing cancer. You have told him that your mother, uncle and grandfather all had cancer, either breast or prostate cancer, when they were around 40 years of age. Your doctor is concerned about your family history and suggests that you see a genetic counselor to learn about genetic testing. The purpose of genetic testing is to see if you have a faulty gene that puts you at high risk for certain types of cancer.

When you visit the genetic counselor she asks you questions about your family's cancer history. Based on this, she says that you qualify for genetic testing. She says that not everyone wants genetic testing because the results can be upsetting. It is up to you to decide if you want genetic testing or not. In the next question you will be asked to make a (hypothetical) decision about undergoing genetic testing. Below is some information about genetic testing to help you with your decision.

What You Could Learn from Genetic Testing

You can get three types of results:

<i>Type of Result</i>	<i>Explanation</i>	<i>Meaning for your Cancer Risk</i>
Positive Result	You have a <u>faulty gene</u>	You are at higher risk get certain cancers in your lifetime than people without a faulty gene. For the faulty gene with the biggest cancer risk, about 8 in 10 people will get cancer. Some cancers you could learn you are at risk for are breast, prostate, pancreatic and ovarian cancers.
Negative Result	<u>No faulty gene</u> was found	You may not have inherited a faulty gene that gives you a high risk to have certain cancers. However, your family history may still put you at higher risk than the general public to get certain cancers. You could still have a faulty gene that was not tested.
Inconclusive Result	You have a change in a gene but it is <u>unclear</u> if this increases your risk of cancer.	An unclear result means that the test cannot help determine your cancer risk at this time. Your family history may still put you at higher risk than the general public to get certain cancers. You could still have a faulty gene that was not tested.

What Would Happen if You Have a Positive Result

If you have a positive result (faulty gene found):

- You will be told about your chance to develop certain cancers.
- Doctors may recommend that you have certain tests to look for cancer (such as a mammogram or PSA testing), or surgeries to prevent cancer.
- There may not be ways to find early and prevent every cancer you could be at high risk for.
- Each of your children would have a 50% chance of having the same faulty gene.
- Other family members may also have the same faulty gene and a high cancer risk.

Logistics

- Genetic testing is done on a tube of your blood.
- There is a 100 dollar copay for any type of genetic testing.

APPENDIX D: Group 1 Genetic Testing Decision Questions

You have the following genetic testing options.

Please pick your preferred option:

No Genetic Testing

5 Gene Test

Description	Looks at the 5 best-studied genes that can cause breast and prostate cancers
Quality of information and recommendations if a positive test	Clear guidelines telling your cancer risks and suggestions for screening
Chance of an inconclusive result	2 in 100 people have an inconclusive result
Time it takes to get your results	3 weeks

>>

Survey Completion
0% 100%

Before the counselor orders the test, she wants you to think about what you would like to learn about your genetic risks for cancer.

Please pick which of the following is more important to you:

- ☐ Having only genetic information with clear meaning for your future health care
- ☐ Having all possible genetic information

>>

Survey Completion
0% 100%

Because you decided to have genetic testing, the counselor would like to let you know of another testing option. This test has 10 additional genes.

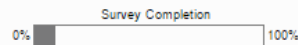
Compared to the 5 gene test, this 15 gene test:

- Tests 10 more genes tied to breast and prostate cancer
 - The health meaning of a positive result (faulty gene found) in these other genes is not well-known
 - 15 out of 100 people have an inconclusive result (versus 2 in 100 for the 5 gene test)
 - The test takes 8 weeks to run (versus 3 weeks for the 5 gene test)
-

Since you said that the meaning of information is more important to you than the amount of information she thinks the 5 gene test is the best option for you. **Would you like to stay with your original answer of the 5 gene test?**

- ☐ Yes, I would like to stay with my original answer of the 5 gene test
- ☐ No, I would like to switch to the 15 gene test
- ☐ No, I don't want genetic testing at all

>>



Because you decided to have genetic testing, the counselor would like to let you know about another testing option. This test has 10 additional genes.


Compared to the 5 gene test, this 15 gene test:

- Tests 10 more genes tied to breast and prostate cancer
 - The health meaning of a positive result (faulty gene found) in these other genes is not well-known
- 15 out of 100 people have an inconclusive result (versus 2 in 100 for the 5 gene test)
- The test takes 8 weeks to run (versus 3 weeks for the 5 gene test)

Since you said that the amount of information is more important to you than the meaning for that information she thinks the 15 gene test is the best option for you. **Would you like to switch to the 15 gene test?**

- ☐ Yes, I would like to switch to the 15 gene test.
- ☐ No, I would like to stay with my original answer of the 5 gene test
- ☐ No, I don't want genetic testing at all

>>

Survey Completion
0%  100%

There is another genetic test that the counselor could have offered you that has 15 genes instead of 5 genes.


Compared to the 5 gene test, this 15 gene test:

- Tests 10 more genes tied to breast and prostate cancer
 - The health meaning of a positive result (faulty gene found) in these other genes is not well-known
- 15 out of 100 people have an inconclusive result (versus 2 in 100 for the 5 gene test)
- The test takes 8 weeks to run (versus 3 weeks for the 5 gene test)

Would you have wanted genetic testing if you had been offered this test?

- ☐ Yes
- ☐ No

>>

Survey Completion
0%  100%

APPENDIX E: Group 2 Genetic Testing Decision Questions

You have the following genetic testing options.

Please pick your preferred option:

	5 Gene Test	15 Gene Test
No Genetic Testing		
Description	Looks at the 5 best-studied genes that can cause breast and prostate cancers	Looks at the 5 best-studied genes that can cause breast and prostate cancers and 10 other genes that have been tied to breast or prostate cancers
Quality of information and recommendations if a positive test	Clear guidelines telling your cancer risks and suggestions for screening	Clear guidelines telling your cancer risks and suggestions for screening for 5 genes. Meaning of a positive result for the 10 other genes is not well-established.
Chance of an inconclusive result	2 in 100 people have an inconclusive result	15 in 100 people have an inconclusive result
Time it takes to get your results	3 weeks	8 weeks

>>

Survey Completion
0% 100%

Before the counselor orders the test, she wants you to think about what you would like to learn about your genetic risks for cancer.

Please pick which of the following is more important to you:

- ☐ Having only genetic information with clear meaning for your future health care
- ☐ Having all possible genetic information

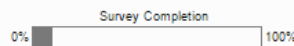
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Survey Completion
0% 100%

Since you said that the meaning of information is more important to you than the amount of information she thinks the 5 gene test is the best option for you. **Would you like to stay with your original answer of the 5 gene test?**

- ☐ Yes, I would like to stay with my original answer of the 5 gene test
- ☐ No, I would like to switch to the 15 gene test
- ☐ No, I don't want genetic testing at all

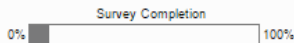
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Since you said that the amount of information is more important to you than the meaning of that information she thinks the 15 gene test is the best option for you. **Would you like to change from your first answer of the 5 gene test to the 15 gene test?**

- ☐ Yes, I would like to switch to the 15 gene test
- ☐ No, I would like to stay with my original answer of the 5 gene test.
- ☐ No, I don't want genetic testing at all

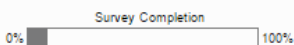
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Since you said that the meaning of information is more important to you than the amount of information she thinks the 5 gene test is the best option for you. **Would you like to change from your first answer of the 15 gene test to the 5 gene test?**

- ☐ Yes, I would like to switch to the 5 gene test
- ☐ No, I would like to stay with my original answer of the 15 gene test.
- ☐ No, I don't want genetic testing at all

>>



Since you said that the amount of information is more important to you than the meaning of that information she thinks the 15 gene test is the best option for you. **Would you like to stay with your original answer of the 15 gene test?**

- ☐ Yes, I would like to stay with my original answer of the 15 gene test.
- ☐ No, I would like to switch to the 5 gene test
- ☐ No, I don't want genetic testing at all

>>



APPENDIX F: Scales and Demographic Measures

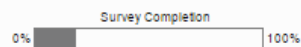
Please answer the following questions about your experience with making the choice of which genetic test to have:

	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
I feel like I understood the medical scenario and genetic testing choice	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I am clear about the best choice for me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I feel sure of what to choose	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This decision is easy for me to make	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I know which options are available to me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I know the pros of each option	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
I know the cons of each option	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I feel I know the benefits of genetic testing	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I know how important the benefits of genetic testing are to me in this decision	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I know how important the cons of genetic testing are to me in this decision	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
It's hard to decide if the benefits of genetic testing are more important to me than the cons	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I feel that others in my life would likely support the choice I have made	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
I have enough advice and information to make a choice	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I feel I have made an informed choice	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
My decision shows what is important to me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I expect to stick with my decision	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I am satisfied with my decision	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I think that I might regret my decision in the future	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree

For each of the following questions, please check the box that best reflects your answer:

	Not at all characteristic of me	A little characteristic of me	Somewhat characteristic of me	Very characteristic of me	Entirely characteristic of me
Unforeseen events upset me greatly.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
It frustrates me not having all the information I need.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Uncertainty keeps me from living a full life.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
One should always look ahead so as to avoid surprises.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
	Not at all characteristic of me	A little characteristic of me	Somewhat characteristic of me	Very characteristic of me	Entirely characteristic of me
A small unforeseen event can spoil everything, even with the best of planning.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
When it's time to act, uncertainty paralyzes me.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
When I am uncertain I can't function very well.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I always want to know what the future has in store for me.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
	Not at all characteristic of me	A little characteristic of me	Somewhat characteristic of me	Very characteristic of me	Entirely characteristic of me
I can't stand being taken by surprise.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
The smallest doubt can stop me from acting.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I should be able to organize everything in advance.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I must get away from all uncertain situations.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
	Not at all characteristic of me	A little characteristic of me	Somewhat characteristic of me	Very characteristic of me	Entirely characteristic of me

>>



For the following questions you will be presented with several words that patients in genetics clinics sometimes struggle with.

Please rate how strongly you agree with the following statement:

I am familiar with the word...

	1- Strongly Disagree	2	3	4	5	6	7- Strongly Agree
Genetic	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Chromosome	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Susceptibility	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Mutation	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Variation	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Abnormality	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Heredity	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Sporadic	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

For the following questions please pick the best word to fill in the blank:

For the following questions please pick the best word to fill in the blank:

Genetics is the study of how living things receive common traits from previous _____.

- ☐ generations
- ☐ expressions
- ☐ partners
- ☐ geminations

A chromosome contains all of our _____ material.

- ☐ genetic
- ☐ digestive
- ☐ cellular
- ☐ brain

Susceptibility to a disease means you _____ get the disease

- ☐ eventually will
- ☐ soon will
- ☐ might
- ☐ will never

A gene mutation is a change in your _____.

- ☐ lipids
- ☐ hair
- ☐ DNA
- ☐ red blood cells

Having a variation in the genetic code will lead to disease _____.

- ☐ all of the time
- ☐ some of the time
- ☐ never
- ☐ only in animals

_____ is an abnormality in humans.

- ☐ a trachea
- ☐ brown hair
- ☐ trisomy
- ☐ blood pressure

Heredity is the transfer of characteristics from _____.

- ☐ the environment to the person
- ☐ the sick to the healthy
- ☐ parent to child
- ☐ teacher to student

A genetic disease that occurs without _____ is considered sporadic.

- ☐ symptoms
- ☐ a family history
- ☐ a diagnosis
- ☐ medication

>>

For each of the following questions, please select the circle that best reflects **how good you are at doing the following things**:

	1- Not at all good	2	3	4	5	6- Extremely Good
How good are you at working with fractions?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How good are you at working with percentages?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How good are you at calculating a 15% tip?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How good are you at figuring out how much a shirt will cost if it is 25% off?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
	1- Not at all good	2	3	4	5	6- Extremely Good

Please check the box that **best reflects your answer**:

	1- Not at all Helpful	2	3	4	5	6- Extremely Helpful
When reading the newspaper, how helpful do you find tables and graphs that are parts of a story?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

Please check the box that **best reflects your answer**:

	1- Always Prefer Words	2	3	4	5	6- Always Prefer Numbers
When people tell you the chance of something happening, do you prefer that they use words ("it rarely happens") or numbers ("there's a 1% chance")?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>


Please check the box that **best reflects your answer:**

	1- Always Prefer Percentages	2	3	4	5	6- Always Prefer Words
When you hear a weather forecast, do you prefer predictions using percentages (e.g., "there will be a 20% chance of rain today") or predictions using only words ("there is a small chance of rain today")?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

Please check the box that **best reflects your answer:**

	1- Never	2	3	4	5	6- Very Often
How often do you find numerical information to be useful?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

>>

Survey Completion
0%  100%

Please answer the following questions about your personal relationship with cancer and your health. (These questions are about you and not the scenario earlier in the survey)

Please indicate if any of the following members of your family have had cancer. **Select all that apply.**

- ☐ Myself
- ☐ Spouse
- ☐ Son
- ☐ Daughter
- ☐ Sister
- ☐ Brother
- ☐ Mother
- ☐ Father
- ☐ Aunt
- ☐ Uncle
- ☐ Grandmother
- ☐ Grandfather
- ☐ Female Cousin
- ☐ Male Cousin
- ☐ Niece
- ☐ Nephew
- ☐ None of the above

In general, how would you rate your overall health?

- ☐ Excellent
- ☐ Very good
- ☐ Good
- ☐ Fair
- ☐ Poor

How do you perceive your cancer risk relative to others of your age and gender?

- ☐ Higher
- ☐ About the same
- ☐ Lower

How would you rate your current level of anxiety about developing cancer?

- ☐ High
- ☐ Moderate
- ☐ Low

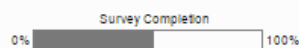
Have you engaged in any of the following cancer screening and prevention behaviors in the past 10 years? **Select all that apply.**

- ☐ Colonoscopy, sigmoidoscopy or fecal occult blood test (FOBT)
- ☐ Endoscopy
- ☐ Digital rectal exam
- ☐ CA-125 blood test
- ☐ Pap test or pelvic exam
- ☐ Transvaginal ultrasound
- ☐ PSA blood test
- ☐ Clinical breast exam
- ☐ Breast self-exam
- ☐ Mammogram, breast MRI or other breast imaging
- ☐ Skin clinical or self-examination
- ☐ Lung CT scan
- ☐ Other (please specify)
- ☐ None

To the best of your knowledge, have you or anyone else in your family ever had cancer genetic testing?

- ☐ Yes
- ☐ No

>>



Please answer the following demographic questions about yourself.

What is your sex?

- ☐ Male
- ☐ Female

How many years old are you?

Please select your age:

What is the highest level of school you have completed or the highest degree you have received?

- ☐ Less than high school degree
- ☐ High school or equivalent
- ☐ Some college
- ☐ Associate or Bachelor degree
- ☐ Graduate degree

What is your annual household income?

- ☐ \$0-\$24,999
- ☐ \$25,000-\$49,999
- ☐ \$50,000-\$99,999
- ☐ \$100,000-\$149,999
- ☐ \$150,000+

How many biological children do you have?

- ☐ 0
- ☐ 1
- ☐ 2
- ☐ 3
- ☐ 4 or more

Which of the following categories best describes your employment status?

- ☐ Employed, working 40 or more hours per week
- ☐ Employed, working 1-39 hours per week
- ☐ Not employed, looking for work
- ☐ Not employed, NOT looking for work
- ☐ Retired
- ☐ Disabled, not able to work

What is your ethnicity?

- ☐ Hispanic or Latino
- ☐ Not Hispanic or Latino

What is your race? (One or more categories may be selected)

- ☐ American Indian or Alaska Native
- ☐ Asian
- ☐ Black or African American
- ☐ Native Hawaiian or Other Pacific Islander
- ☐ White

Is English your native language?

- ☐ Yes
- ☐ No

Which state do you currently live in?

Please select your state from the drop-down menu

How would you describe your political beliefs?

- ☐ Liberal
- ☐ Conservative
- ☐ Independent
- ☐ Other (please specify)

Which of the following best describes your current relationship status?

- ☐ Married
- ☐ Widowed
- ☐ Divorced
- ☐ Separated
- ☐ In a domestic partnership or civil union
- ☐ Single, but cohabiting with a significant other
- ☐ Single, never married

Please enter your preferred email address so that we can send you the e-gift card.

>>

Survey Completion
0% 100%

APPENDIX D: Gift Card Distribution Message

Hello,

Thank you for participating in my study, *Decisions about Genetic Testing for Hereditary Cancer Predisposition*. Below is your Amazon.com gift card!

Thanks,
Marci Barr

e-gift card displayed here

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CURRICULUM VITAE

Marci Barr

EDUCATION

JOHNS HOPKINS UNIVERSITY, NATIONAL HUMAN GENOME RESEARCH INSTITUTE-NIH

Sc.M IN GENETIC COUNSELING
Expected 2015 | Baltimore, MD
Cum. GPA: 3.94 / 4.0

FRANKLIN AND MARSHALL COLLEGE

BA IN BIOLOGY

Grad May 2011 | Lancaster, PA
Cum. GPA: 3.76 / 4.0
Magna Cum Laude
Class Rank: 46/540
Phi Beta Kappa Honor Society
Marshall Scholar
Courtney Adams Music Scholarship

UNIVERSITY OF OTAGO

UNDERGRADUATE STUDY ABROAD
Spring 2010 | Dunedin, New Zealand
Nominee, Best Chindogu for Invention "sit
anywhere anytime pants" at Dunedin
Fringe Festival
Awarded "Most Creative" by Institute for
Study Abroad staff

AWARDS

2014 | Best Student Abstract | *National
Society of Genetic Counselors
Annual Education Conference*
2010 | Nolt Music Award | *University
Musical Transmission and the Hammered
Dulcimer*
2010 | Paul A. Mueller Jr. Summer Travel
Award | *University*
Yoga Instructor Certification
2005 | Girl Scout Gold Award | *National
The Prairie Warbler Trail: Environmental
Conservation and Community Education
Girl Scouts of the USA*

SKILLS

COMPUTER

Microsoft Office
Stata, PASW Statistics
L^AT_EX
Windows and Macintosh operating
systems

LANGUAGE

English (Fluent)
Spanish, German(Basic Knowledge)

GENETIC COUNSELING ROTATION EXPERIENCE

THE JOHNS HOPKINS HOSPITAL-CLINICAL CANCER GENETICS

| BREAST/OVARIAN, COLORECTAL, AND FAMILIAL CANCERS
September-December | Baltimore, MD

NATIONAL HUMAN GENOME RESEARCH INSTITUTE, NIH

| RESEARCH INTERN- OVERGROWTH DISORDERS AND MMA
June-July 2014 | Bethesda, MD

THE JOHNS HOPKINS HOSPITAL-PEDIATRIC GENETICS

| GENERAL, SKELETAL DYSPLASIAS, OCULAR, CONNECTIVE TISSUE
March-May 2014 | Baltimore, MD

THE JOHNS HOPKINS HOSPITAL- ADULT DISORDERS

| GENERAL INTERNAL MEDICINE AND CONNECTIVE TISSUE DISORDERS
October-December 2013 | Baltimore, MD

GREATER BALTIMORE MEDICAL CENTER- HARVEY INSTITUTE FOR HUMAN GENETICS | CANCER AND EDS

September-October 2013

GENOMES2PEOPLE RESEARCH GROUP-BRIGHAM AND WOMEN'S HOSPITAL | RESEARCH INTERN

June - July 2013 | Boston, MA
Assisted projected managers on three translational genomics projects

WALTER REED NATIONAL MILITARY MEDICAL CENTER

| PRENATAL GENETICS
January- May 2013 | Bethesda, MD

GENETIC AND RARE DISEASES INFORMATION CENTER

| INFORMATION SPECIALIST INTERN
October- December 2012 | Gaithersburg, MD
Answered telephone and written inquiries about genetic and rare diseases.

WORK EXPERIENCE

TEACHING ASSISTANT | INTRODUCTION TO BIOETHICS IN PUBLIC HEALTH PRACTICE AND RESEARCH

July 2014 | Baltimore, MD
• Grading TA, Johns Hopkins University Bloomberg School of Public Health.

NATIONAL HEART LUNG AND BLOOD INSTITUTE, NIH

| POST-BACCALAUREATE INTRAMURAL RESEARCH TRAINING Awardee
September 2011 - August 2012 | Bethesda, MD

- Primary Project: Evaluation of preferences during informed consent for genetic research in individuals with sickle cell disease
- Auxiliary Projects: Admixture mapping and microarray studies to investigate genetic modifiers of pain in sickle cell disease

EASTON HOSPITAL | INTERNSHIP AND LABORATORY SERVICE REPRESENTATIVE

June 2009 - September 2011 | Easton, PA

- Answering inquiries of hospital staff and patients
- Contacting patients and offices to clarify and correct information
- Preparing cytology and microbiology specimens for analysis

COURSEWORK

GRADUATE

Human Genetics
Counseling Skills
Research
Public Health
Clinical Rotation Experience

UNDERGRADUATE

Biology
Chemistry
Physics and Calculus
Music
Liberal Arts Courses

EXTRACURRICULAR

UNDERGRADUATE

Principle Oboist | Franklin and Marshall
Orchestra and Philharmonia
Data Manager | Sustainability Theme
House
Member and Fair Trade Cafe Volunteer |
Environmental Action Alliance
Substitute Yoga Instructor and
Participant | On-Campus Yoga
Hospital Shadowing | Lancaster General
Hospital Medical Preceptorship Program
Representative 1st Dubbs Hallway |
Boncheck College House Congress
Co-organizer campus-wide clothing swap
| Kappa Delta Sorority

GRADUATE

Oboist | NIH Community Orchestra
Student Interviewer | Genetic Counseling
Associate Director Selection Committee
Participant | Group Fitness Classes

WORK EXPERIENCE, CONTINUED

FRANKLIN AND MARSHALL COLLEGE PHONATHON | STUDENT CALLER

January - May 2011 | Lancaster, PA

- Informed alumni about college current events
- Established Rapport with Alumni
- Asked alumni for contributions to the College Annual Fund

GIRL SCOUTS OF EASTERN PENNSYLVANIA | COUNSELOR, CAMP MOSEY WOOD

June - December 2008, July-August 2011 | White Haven, PA

- Taught basic outdoors skills, crafts, and games
- Planned and led trips, cookouts, and activities for campers
- Mediated disputes between campers, managed homesickness, and treated minor injuries

VOLUNTEER EXPERIENCE

TURNING POINT OF THE LEHIGH VALLEY | DOMESTIC VIOLENCE SHELTER OFFICE AND CRISIS HOTLINE VOLUNTEER

June - August 2011 | Allentown, PA

- Completed 40 hour domestic violence training
- Interacted with shelter residents and helped with the children's support group
- Answered the crisis hotline
- Helped with routine office responsibilities

LA HESPERIA BIOLOGICAL RESERVE | FULL-TIME VOLUNTEER-FUNDED BY MARSHALL AWARD, FRANKLIN AND MARSHALL COLLEGE

September - December 2010 | La Esperie, Ecuador

- Collected and planted seeds from endangered hardwood species
- Maintained gardens and facilities
- Tested research techniques and taught them to research groups
- Conversated with locals and utilized Spanish language skills to travel effectively

GOSCHENHOPPEN FOLK FESTIVAL | VOLUNTEER-DEMONSTRATING 19TH CENTURY CROCHETED LACE EDGING

Annual participation since 1992 | Green Lane, PA

- Demonstrate crocheted lace edging
- Explain the historical significance of my craft in Pennsylvania German culture to festival visitors

PRESENTATIONS

2014 | *Genetic Counselors as Choice Architects: Some Considerations for Presenting Genetic Testing Decisions in a Complex Choice Environment* | Best Student Abstract Plenary, National Society of Genetic Counselors Annual Education Conference

2013 | *Preferences and Understanding: Informed Consent for Genetic Research among Individuals with Sickle Cell Disease* | National Human Genome Research Institute Retreat, National Institutes of Health

2012 | *Understanding and Attitudes: Informed Consent of Patients with Hemoglobinopathies for Genetic Research* | Post-baccalaureate Research Symposium, National Institutes of Health

2011 | *Integration of Forest Conservation and Sustainable Agriculture Practices in Ecuador* | Franklin and Marshall College Spring Research Fair

2011 | *Musical Transmission and the Hammered Dulcimer* | Franklin and Marshall College Spring Research Fair